INCLUDING CHILDREN AND ADOLESCENTS WITH AUTISM SPECTRUM DISORDER IN FUNCTIONAL BEHAVIOURAL ASSESSMENT-INFORMED INTERVENTIONS FOR SLEEP DISTURBANCE

Thesis submitted in fulfilment of

the requirements for the degree of Doctor of Philosophy in Psychology

by Jenna van Deurs

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Ehara taku toa i te toa takitahi Engari, he toa takitini

Success is not the work of one, but the work of many

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“Don’t think I’m dumb

If your words seem foreign . . .

I’m only human

I’m like you.

I just think different.

Don’t look at me like I’m a child.

Your words may seem foreign

But I’m smarter than you think.

So don’t cry for me.

I am what I am.

I’m special, just like you.”

(Janet Weedon-Skinner, 17-year-old with Autism Spectrum Disorder; Chilvers, 2007, p. 18)

Abstract

Sleep disturbances are a significant problem for individuals with autism spectrum disorder (ASD) across the lifespan and are associated with numerous detrimental impacts to the individual and their whānau/family. The aetiology of ASD-related sleep problems is complex and likely multifactorial. Functional behavioural assessment (FBA) is increasingly being utilised to identify environment-behaviour contingencies underlying sleep disturbance and inform targeted treatment. Parent-implemented behavioural sleep interventions for young children with ASD are well supported by research, but few studies have evaluated such interventions for older children and adolescents, nor has research investigated the feasibility or effectiveness of young person-implemented interventions. Using single-case experimental designs (multiple-baseline design or AB design with replication across participants and behaviours), the three empirical studies included in this thesis evaluated the efficacy of FBA-informed parent- and young person-implemented sleep interventions with 12 verbally communicative 9- to 15-year-olds with ASD (11 males, 1 female). The results illustrated intervention was generally effective in reducing or resolving participant sleep disturbance and improvements tended to be maintained at short- and long-term follow-up. Further, the secondary outcomes of resolving sleep disturbance appeared to include a significant reduction in participant internalising (e.g., anxiety, depressive symptoms) behaviour, without significant improvements to participant externalising behaviour, or in parent sleep, mental health, or marital relationship quality. Overall, findings indicate the feasibility, effectiveness, and acceptability of FBA-informed interventions involving input from the young person and their parents.

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K France and L McLay: assisted with the development and conceptualisation of the study, provided clinical supervision, edited the manuscript

N Blampied: constructed the figures, assisted with the selection and understanding of data analysis methods, and edited the manuscript

R Lang: assisted with editing the manuscript

J Hunter: assisted with the assessment & intervention of one participant

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The candidate (80%):

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List of Abbreviations

Adolescent Sleep Hygiene Scale	ASHS
Adolescent Sleep Wake Scale Revised	ASWS-R
American Psychiatric Association	APA
Attention-deficit/hyperactivity disorder	ADHD
Auditory processing disorder	APD
Autism spectrum disorder	ASD
Behavioural Intentions Questionnaire	BIQ
Child Behavior Checklist for Ages 6-18	CBCL
Children's Sleep Habits Questionnaire	CSHQ
Cognitive behavioural therapy	CBT
Cognitive behavioural therapy for insomnia	CBT-I
Common Language Effect Size	CLES
Confidence interval	CI
Curtain calls	CCs
Depression Anxiety and Stress Scale-21	DASS-21
Diagnostic and Statistical Manual of Mental Disorders	DSM
Early wakings	EWs
Functional behaviour assessment	FBA
Generalised anxiety disorder	GAD
Gilliam Autism Rating Scale - Third Edition GARS-3	GARS-3

High functioning autism	HFA
Intellectual disability	ID
Interobserver agreement	IOA
Multidimensional Anxiety Scale for Children 2nd Edition	MASC-2
New Zealand	NZ
Night wakings	NWs
Nonrapid eye movement	NREM
Polysomnography	PSG
Post-extinction response burst	PERB
Percentage below the median	PBM
Percentage exceeding the median	PEM
Pervasive developmental disorder	PDD
Pervasive developmental disorder- not otherwise specified	PDD-NOS
Pittsburgh Sleep Quality Index	PSQI
Probability of Superiority	PS
Progressive muscle relaxation	PMR
Questions About Behavioral Function	QABF
Rapid eye movement	REM
Relationship Quality Index	RQI
Reliable change index	RCI
Sleep Assessment and Treatment Tool	SATT

Sleep efficiency	SE
Sleep onset latency	SOL
The Children's Sleep Comic	CSC
The Sleep Self-Report	SSR
Theory of mind	TOM
Treatment Acceptability Rating Form-Revised	TARF-R
United Nations	UN
Video modelling	VM
Video self-modelling	VSM
Videosomnography	VSG
Vineland Adaptive Behavior Scales, Second Edition	VABS-II
Vineland Adaptive Behavior Scales, Third Edition	Vineland-3
Young Person Treatment Evaluation	YPTE
Wake-after-sleep-onset	WASO

Chapter 1

Introduction

Autism Spectrum Disorder

Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterised by social communication difficulties and engagement in restricted or repetitive patterns of behaviour, interests, and activities (American Psychiatric Association [APA], 2013). In addition to these core features, speech and language ability may also be affected. This can manifest as delays in the development of spoken language, difficulty engaging in and understanding non-verbal communication, and lack of intelligible speech. The sensory processing of individuals with ASD is also commonly affected, contributing to hypo or hyper reactions to stimuli. Autism is a heterogenous disorder and there is wide variation in the extent to which affected individuals experience the preceding challenges. Individuals on the spectrum may experience mild symptom severity with low impact on their cognitive and adaptive functioning, while others may be classified as having severe deficits within affected areas, requiring substantial ongoing support (APA, 2013).

Autism was first described by Kanner (1943), based on the behaviour of 11 children who experienced difficulty relating to others, demonstrated a lack of interest in people and socialisation, had atypical language, and engaged in stereotyped behaviour. In 1980 autism became an officially recognised diagnosis in the third edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-*III*; Volkmar et al., 2014). Prior to publication of the fifth edition of the DSM (DSM-5) in 2013, autism was considered to be a pervasive developmental disorder (PDD), alongside Rett's disorder, Asperger's disorder, childhood disintegrative disorder, and pervasive developmental disorder-not otherwise specified (PDD-NOS; Volkmar et al., 2014). Individuals diagnosed with Rett's disorder experienced decelerated head growth, loss of purposeful hand skills, increased stereotyped hand movements, loss of social participation, as well as impaired language and psychomotor abilities (APA, 2000; Van Acker, Loncola, & Van Acker, 2005). Childhood disintegrative disorder referred to children who experienced regression in key areas of development (e.g., motor skills) and demonstrated challenges related to social interaction, communication, or stereotyped movements (APA, 2000). Individuals diagnosed with Asperger's disorder were differentiated from people with autism based on the lack of language impairment and higher intellectual ability (APA, 2000; Klin, McPartland, & Volkmar, 2005). Individuals diagnosed with Asperger's displayed subtle differences in social

communication, for example, using meticulous speech, or frequently engaging in specific topics of interest (APA, 2000; Volkmar et al., 2014). Those who did not meet criteria for a PDD, but experienced mild symptoms or difficulties in select areas relating to social communication or restricted/repetitive behaviours, were diagnosed with PDD-NOS (APA, 2000; Towbin, 2005).

In 2013, in accordance with the DSM-5, all PDDs were subsumed into one category, ASD (Volkmar et al., 2014). This decision reflected the lack of evidence of definitive and replicable differences between diagnostic categories (Volkmar et al., 2014). In contrast with the fourth edition of the DSM (DSM-IV-TR), the DSM-5 includes specifiers such as, language impairment and intellectual impairment, to increase descriptive subtyping of individuals previously diagnosed with a PDD (Volkmar et al., 2014). Additionally, the following terms can be used to indicate the level of support required: "Requiring support", "Requiring substantial support", and "Requiring very substantial support". Participants within this thesis include those diagnosed with ASD as well as individuals diagnosed with a PDD prior to 2013.

Historically, people on the autism spectrum were classified as 'high' or 'low functioning'. High functioning autism (HFA) referred to individuals without a comorbid intellectual disability (ID), or an intelligence quotient (IQ) above 70 (Richdale, Baker, Short, & Gradisar, 2014). These individuals tended to have more developed functional language skills and were more likely to be diagnosed with Asperger's prior to the DSM changes (Carr, Moore, & Anderson, 2014). However, the terms high and low functioning are generally not preferred by individuals on the autism spectrum or their family members (Kenny et al., 2016). Consequently, participants within the current thesis will not be categorised as high or low functioning.

As yet, no research has investigated the prevalence of ASD in New Zealand (NZ). However, based on prevalence rates within the United Kingdom, the Ministries of Health and Education (2016) estimate 1 in 100 people in NZ are on the autism spectrum. Given the current population of NZ, this amounts to approximately 50,000 people. Interestingly, males are more commonly diagnosed with ASD than females, with a male-to-female diagnostic ratio of 4:1 (Loomes, Hull, & Mandy, 2017). This difference is purported to be linked to diverse symptom presentations (i.e., increased internalising behaviour in females versus externalising behaviour in males) and subtler differences between females with mild to moderate symptoms and those who are typically developing (Attwood, 2007; Lai, Lombardo, Auyeung, Chakrabarti, & Baron-Cohen, 2015; Loomes et al., 2017). Thus, diagnostic criteria may align better with male ASD phenotypes (Loomes et al., 2017).

Sleep Disturbance in ASD

Although it is not a diagnostic criterion of the disorder, sleep disturbance is a common co-occurring issue for people with ASD (Allik, Larsson, & Smedjie, 2006; Elrod & Hood, 2015; Elrod et al., 2016; Malow, Katz et al., 2016; Meltzer, Johnson, Crosette, Ramos, & Mindell, 2010; Ming, Brimacombe, Chaaban, Zimmerman-Bier, & Wagner, 2008; Polimeni, Richdale, & Francis, 2005; Williams, Sears, & Allard, 2004). Estimates based on recent large-scale studies (n > 1,000) suggest prevalence rates in children and adolescents with ASD are between 31 to 74% (Elrod et al., 2016; Goldman, Richdale, Clemons, & Malow, 2012; Malow, Katz et al., 2016), compared with just 9 to 50% in typically developing young people (Goldman et al., 2012; Kotagal & Broomall, 2012). Further, children with ASD may experience higher rates of sleep problems than children with other developmental disorders (Cotton & Richdale, 2006). Despite the wide variation in reported rates (likely stemming from diverse definitions and sleep measurements used), it is clear sleep disturbance is a significant issue for young people with ASD (Allik et al., 2006; Allik, Larsson, & Smedjie, 2008; Hodge, Carollo, Lewin, Hoffman & Sweeney, 2014; Paavonen et al., 2008; Park et al., 2012; Richdale, 2013; Richdale & Baker, 2014; Souders et al., 2009). Currently, there is a distinct lack of data in relation to the rates of sleep disturbance across the spectrum, although most studies reveal symptom severity does not predict sleep problem severity (Krakowiak, Goodlin-Jones, Hertz-Picciotto, Croen, & Hansen, 2008; Patzold, Richdale, & Tonge, 1998; Polimeni et al., 2005; Williams et al., 2004).

Dyssomnias are a broad category of sleep disorders relating to difficulty establishing and maintaining sleep (e.g., insomnia, obstructive sleep apnoea), or excessive sleepiness (e.g., hypersomnolence). Insomnia is the most common form of sleep disturbance experienced by children and adolescents with ASD (Cortesi, Giannotti, Ivanenko, & Johnson, 2010; Goldman et al., 2012; Loring, Johnston, Gray, Goldman & Malow, 2016) and is the focus of the current research. Insomnia may manifest as abnormal sleep-wake patterns, poor sleep efficiency (SE; percentage of time spent asleep in bed relative to total time in bed), delayed sleep onset latency (SOL; time taken to fall asleep once in bed), frequent and extended night wakings (NWs), or early morning waking (EW; any rise time before 6:00am where sleep is not reinitiated). Such sleep problems are often associated with bedtime resistance (e.g., crying, curtain calls [CCs]; bids for parental attention from the young person post-bedtime prior to sleep initiation]), problematic NW behaviours (e.g., wandering through the house), and unwanted co-sleeping (Kirkpatrick, Gilroy, & Leader, 2019; Richdale, 2013). Parasomnias refer to behaviours or physiological events which occur while an individual is asleep (e.g., night terrors, nightmares,

sleep walking/talking, bruxism). Similarly to typically developing young people, parasomnias are more common among very young children with ASD (Goldman et al., 2012), although, overall, individuals with ASD are more likely to present for help with dyssomnias than parasomnias. The assessment and treatment requirements of parasomnia fall outside the aims of this research. Consequently, parasomnias are not examined in the current study.

Implications of Sleep Disturbance

Numerous bidirectional associations are apparent between sleep and a range of cognitive, behavioural, psychological, physical, and family factors (Allik et al., 2006; Bourgeron, 2007; Cohen, Conduit, Lockley, Rajaratnam & Cornish, 2014; Cortesi et al., 2010; Goldman et al., 2012; Herrmann, 2016; Jin, Hanley, & Beaulieu, 2013; Kotagal & Broomall, 2012; May, Cornish, Conduit, Rajaratnam & Rinehart, 2015; Malow et al., 2014; Moon, Corkum, & Smith, 2011; Park et al., 2012; Richdale & Schreck, 2009; Richdale & Wiggs, 2005; Souders et al., 2009; Taylor, Schreck, & Mullick, 2012; Tilford et al., 2015; Turner & Johnson, 2013; Vriend, Corkum, Moon, & Smith, 2011). Notably, sleep disturbance is linked to detrimental outcomes in each of the preceding areas.

Cognitive outcomes. Sleep is thought to play a critical role in establishing and maintaining effective cognitive functioning (Taylor et al., 2012). Sleep is implicated directly in brain maturation, executive functioning, memory consolidation, information processing, and learning (Turner & Johnson, 2013). Sufficient sleep is required for events or procedures to be encoded within long-term memory, particularly cognitive and motor skills (Kotagal & Broomall, 2012). Accordingly, adequate sleep is necessary to facilitate language acquisition and consolidation (Touchette et al., 2007). Consequently, the concerted efforts of educators, therapists, and caregivers to enhance the skills of young people with ASD are compromised if they do not receive enough sleep. Research with typically developing children illustrates just 30 minutes of sleep loss (from their typical, optimal sleep duration) can lead to neuropsychological impairment (Richdale & Wiggs, 2005). Unsurprisingly, sleep problems have been linked to lower intellectual functioning as well as reduced academic performance in young people with ASD (Bruni et al., 2007; Hollway & Aman, 2011; Giannotti et al., 2008; Owens, Spirito, & McGuinn, 2000; Richdale & Wiggs, 2005; Taylor et al., 2012; Yang et al., 2018).

Behavioural outcomes. Numerous studies have identified an association between sleep disturbance and exacerbated autism symptomology (Allik et al., 2006; Cohen et al., 2014; Cortesi et al., 2010; Goldman et al., 2011; Hoffman et al., 2005; Hundley, Shui & Malow, 2016;

Schreck, Mulick, & Smith, 2004), including poorer social communication skills and social interactions (Elia et al., 2000; Malow, McGrew, Harvey, Henderson, & Stone, 2006; Goldman et al., 2011; Schreck et al., 2004; Taylor et al., 2012; Yang et al., 2018). In fact, Phung and Goldberg (2017) found social communication difficulties were intensified in adolescents with ASD and sleep disturbance, whereas this was not the case for typically developing individuals with sleep problems. Phung and Goldberg (2017) suggest typically developing young people may be better able to regulate their social interactions in spite of sleep deprivation, whereas existing social communication deficits are likely further compromised amongst adolescents with ASD. Parents of children with ASD and insomnia have attributed their child's lack of social interaction to exhaustion (Kirkpatrick et al., 2019). In addition to social communication difficulties, sensory processing issues and stereotypic or repetitive behaviour during the day is also intensified amongst young people with ASD and sleep problems (Abel, Schwichtenberg, Brodhead, & Christ, 2018; Hundley et al., 2016; Goldman, Surdyka, Cuevas, Adkins, Wang, & Malow, 2009; Mazurek & Petroski, 2015; Reynolds, Lane, & Thacker, 2015; Schreck et al., 2003; Tyagi, Juneja, & Jain, 2019).

A number of studies also indicate that sleep problems may exacerbate externalising behaviour problems in young people with ASD, including aggression, hyperactivity, noncompliance, and self-injurious behaviour (Adams, Matson, & Jang, 2014; Allik et al., 2006; Bruni et al., 2007; Cohen et al., 2014; Goldman et al., 2011; Mayes & Calhoun, 2009; Mazurek & Sohl, 2016; Park et al., 2012; Patzold et al., 1998; Sikora, Johnson, Clemons, & Katz, 2012; Soke et al., 2017). This is consistent with research investigating the impact of sleep disturbance on typically developing young people or individuals with an ID (Gregory & Sadeh, 2012; Richdale, Francis, Gavidia-Payne, & Cotton, 2000). Inadequate sleep is thought to impair impulse control, likely predisposing individuals with poor sleep to engage in externalising behaviour (Cohen et al., 2014).

The adaptive functioning of young people with ASD is also thought to be affected by sleep disturbance (Sikora et al., 2012; Taylor et al., 2012). For example, young people who receive less sleep have greater difficulty completing daily living tasks (e.g., eating, brushing hair, toileting; Taylor et al., 2012; Tyagi, Juneja, & Jain, 2019). Tyagi et al. (2019) propose impairments related to sleep disturbance (e.g., fatigue, inattention, memory loss) may inhibit skill acquisition.

Psychological outcomes. Internalising behaviour is also related to sleep quality (Allik et al., 2006; Hollway, Aman, & Butter, 2013; Mazurek & Petroski, 2015; Richdale & Baglins, 2015; Tani et al., 2003). Young people with ASD and sleep disturbance are more likely to demonstrate negative affect, and experience depressed moods, anxiety, and increased emotionality (Abel et al., 2018; Allik et al., 2006; Goldman et al., 2011; Nadeau et al., 2015; Mayes & Calhoun, 2009; Mazurek & Petroski, 2014; Richdale & Baglin, 2015; Richdale & Wiggs, 2005; Rzepecka, McKenzie, McClure, & Murphy, 2011; Tani et al., 2003; Uren, Richdale, Cotton, & Whitehouse, 2019; Wiggs & Stores, 2004). Although anxiety and depressive psychopathology is common among people with ASD, it is more frequent among those with comorbid sleep problems (Mazzone, Postorino, Siracusano, Riccioni, & Curatolo, 2018; Uren et al., 2019). Inadequate sleep is thought to compromise emotional, behavioural, and cognitive regulation, likely contributing to and maintaining psychological issues (Kotagal & Broomall, 2008; Vandekerckhove & Cluydts, 2010).

Physical outcomes. Not only does sleep disturbance have implications for psychosocial wellbeing, but it can also affect physical wellbeing. Delahaye et al. (2014) found a strong relationship between sleep problems in older children with ASD and poor physical health-related quality of life. Further, sleep problems have also been linked to gastrointestinal problems, epilepsy, obesity, and respiratory issues (Accardo & Malow, 2015; Mannion & Leader, 2013; Williams et al., 2004; Yang et al., 2018; Zuckerman, Hill, Guion, Voltolina, & Fombonne, 2014). Although initial findings suggest physical health and sleep are connected, this area has not been well studied in individuals with ASD.

Family outcomes. The impact of sleep disturbance is not isolated to the affected individual. In fact, it can have detrimental implications for the whole family. Sleep disturbance adds to the significant challenges and stressors already posed by parenting a child with a developmental disability (Goldman et al., 2011; Kodak & Piazza, 2008). In addition, the sleep quality of other household members is often reduced (Kirkpatrick et al., 2019; Meltzer, 2008; Moss, Gordon, & O'Connell, 2014; Lopez-Wagner, Hoffman, Sweeney, Hodge, & Gilliam, 2008; Patzold et al., 1998). When describing his experience of fathering a child with ASD and sleep disturbance, Peter (2006) said "fatigue is the currency of daily living. . . For many years I had trouble staying awake at work. Even today I can't sit through a meeting without nodding into sleep" (p.88). In Kirkpatrick and colleagues' (2019) qualitative study investigating the family implications of child insomnia, parents cited financial burden as they were unable to attend work due to their own sleep deprivation.

Child sleep problems have also been shown to contribute to poor parent mental health and marital discord (Durand & Mindell, 1990; Kodak & Piazza, 2008; Kotagal & Broomall, 2012; Levin & Scher, 2016; Lopez-Wagner et al., 2008; Meltzer, 2011; Turner & Johnson, 2013; Vriend et al., 2011). Parent report suggests sleeping separately from one's partner (as a result of co-sleeping) can contribute to relationship deterioration (Kirkpatrick et al., 2019). Further, tension can arise when only one parent is responsible for managing their child's sleep disturbance (Kirkpatrick et al., 2019). Child sleep problems are thought to contribute to emotional distress (e.g., sense of overwhelm) and reduce capacity for enjoyable activities in the day and evening (Kirkpatrick et al., 2019).

Evidently, the high prevalence of sleep problems amongst young people with ASD, in conjunction with neurodevelopmental deficits, further compromises their functioning and exacerbates existing difficulties (e.g., with cognitive, verbal, and motor skills). Given the clear association between inadequate sleep and poor wellbeing, targeted sleep interventions may benefit the quality of life of the affected individual and their whānau/family (Richdale & Wiggs, 2005). Accordingly, research suggests improving sleep disturbance can reduce autism symptom severity, internalising behaviour, externalising behaviour, parent stress, and improve social communication (Loring, Johnston, Shui, & Malow, 2018; Malow et al., 2006; Malow, Adkins et al., 2012; May et al., 2015; Reed et al., 2009).

Aetiology of Sleep Disturbance in ASD

As discussed above, sleep disturbance can contribute to numerous biopsychosocial issues for individuals affected by ASD. However, this relationship is not unidimensional. In fact, the presence of biopsychosocial issues in the first place can precipitate the onset of sleep problems and contribute to their maintenance. Thus, causal factors underlying sleep problems experienced by individuals with ASD are multifaceted and complex. Physiological, environmental, cognitive, and behavioural aetiologies to the problem have been investigated.

Physiological contributions. Individuals with ASD are thought to have disrupted sleep architecture, whereby the typical cyclical organisation of sleep phases throughout the night is altered (Baglioni et al., 2016; Mazzone et al., 2018). There are two distinct phases of sleep, nonrapid eye movement (NREM) and rapid eye movement (REM) sleep. Each of these phases is characterised by certain physiological and behavioural features. For example, there is relatively low brainwave activity during NREM sleep and high brainwave activity during REM sleep (Lushington, Pamula, Martin, & Kennedy, 2013). Typical sleep architecture involves alternating

between NREM and REM sleep phases in approximately 90 to 110 min cycles throughout the night (Mindell & Owens, 2015). NREM sleep is comprised of three stages, each of which consist of progressively deeper sleep and higher arousal thresholds (i.e., stage one is the lightest sleep phase and easiest to wake from, whereas stage three is the deepest sleep phase and most difficult to rouse from; Lushington et al., 2013; Shakankiry, 2011). Children and adolescents typically enter NREM sleep first (Lushington et al., 2013). This slow wave sleep predominates for the first third of the night, before declining as REM sleep increases during the latter part of the night (Mindell & Owens, 2015; Shakankiry, 2011). Both phases are necessary to achieve optimum benefits from sleep. Although the investigation of sleep architecture in individuals with ASD is in its infancy, preliminary research suggests people with ASD may have less REM sleep, less stage three NREM sleep (deep sleep phase), increased stage one NREM sleep (light sleep phase), and slower cyclic alternating patterns between phases (Baglioni et al., 2016; Bourgeron, 2007; Diomedi et al., 1999; Elia et al., 2000; Harvey & Kennedy, 2002; Limoges, Mottron, Bolduc, Berthiaume, & Godbout, 2005; Mazzone et al., 2018; Miano et al., 2007; Richdale & Shreck, 2009). Additionally, differences in sleep phases do not seem to be as pronounced in individuals with ASD; for example, REM sleep consists of higher muscle activity than is typical, comparable with NREM sleep stages (Souders et al., 2017). All of the preceding factors contribute to individuals with ASD having reduced sleep duration and/or sleep quality (Herrmann, 2016).

The physiological profiles of individuals with ASD may also include dysregulated levels of melatonin leading to disrupted circadian rhythms (Glickman, 2010; Melke et al., 2008; Souders et al., 2009; Tordjman, Anderson, Pichard, Charbury, & Touitou, 2005; Tordjman et al., 2012). Melatonin is crucial to regulation of the sleep/wake cycle, helping set one's internal clock to the natural 24-hour day/night cycle (Bourgeron, 2007). Elevated nocturnal melatonin is believed to be implicated in establishing and maintaining sleep (Cortesi et al., 2010; Glickman, 2010; Tordjman et al., 2005; Tordjman et al., 2012). Consequently, dysregulated melatonin levels can disrupt one's ability to initiate sleep at appropriate times. Compared to those without ASD, individuals with ASD are more likely to experience reduced melatonin secretion overall, as well as delayed onset of endogenous melatonin, resulting in elevated melatonin levels during the daytime and reductions at night (Melke et al., 2008; Souders et al., 2009; Tordjman et al., 2005; Tordjman et al., 2012). Subsequent circadian abnormalities may result in children with ASD having different sleep schedules to their family, potentially enhancing resistance and behavioural problems at bedtime (Turner & Johnson, 2013). Further, the most common sleep

disturbances in children with ASD, such as delayed sleep onset and frequent NWs, are consistent with a circadian disturbance (Glickman, 2010).

Characteristics of ASD. Key characteristics of individuals with ASD, including hypersensitivity to environmental stimuli, insistence on routines and sameness, communication difficulties, and difficulty interpreting social cues may also be implicated in their high rates of sleep disturbance (Brown et al., 2014; Loring et al., 2016). Individuals with ASD are more likely to experience sensory modulation difficulties as a result of either a low or high neuronal threshold (Reynolds, Lane, & Thacker, 2012). A low neuronal threshold results in greater sensitivity to sensory stimuli, whereas individuals with a high neuronal threshold require greater sensory stimulation levels to obtain an optimal arousal level (Reynolds et al., 2012). Both of these processes may make young people with ASD more prone to developing sleep disturbance. It is a more effortful process for individuals with a low neuronal threshold to disengage from the sensory environment (Reynolds et al., 2012). Consequently, some young people with ASD may struggle to lower their arousal in response to the absence or presence of certain sounds, sights and feelings, or experience difficulty tuning out environmental stimuli (Mazurek & Petroski, 2014). Conversely, those with a high neuronal threshold may be more likely to engage in sleep-interfering behaviour to seek optimal sensory arousal.

Restricted/repetitive behaviours are a core feature of ASD and can manifest as cognitive inflexibility, insistence on sameness, and ritualistic behaviours (Mazzone et al., 2018). These symptoms may also contribute to the high prevalence of sleep problems in individuals with ASD. For example, such individuals may become distressed if their bedtime routine is altered, have difficulty transitioning between activities in the lead up to bed, and engage in time-consuming rituals pre- and post-bedtime (Mazzone et al., 2018).

Further, people with ASD, particularly those who are non-verbal or have an ID, may struggle to interpret social and environmental cues that bedtime is approaching, as well as how to fall asleep (Brown et al., 2014; Kotagal & Broomall, 2012). Difficulty interpreting social stimuli, can lead to individuals being less aware of, or motivated by social constructs related to sleep, such as an maintaining an appropriate bedtime (Loring et al., 2016). Accordingly, individuals with ASD may have reduced understanding of family sleep expectations (Loring et al., 2016). In addition, reduced sensitivity to social cues may affect the ability of individuals with ASD to synchronise their circadian rhythm (internal biological clock) to external time cues

(e.g., mealtimes), thus leading to misalignment between their circadian phase and external environment (Cohen et al., 2014).

Comorbid conditions. Common comorbid medical and psychological conditions in young people with ASD can also interfere with their sleep (Cohen et al., 2014; Mazzone et al., 2018; Trickett, Heald, Oliver, & Richards, 2018). These comorbidities include: anxiety, depression, attention-deficit/ hyperactivity disorder (ADHD), epilepsy/ seizures, iron deficiency, restless legs syndrome, gastrointestinal problems, allergies, and asthma (Ameis et al., 2018; Cohen et al., 2014; Cortesi et al., 2010; Dosman et al., 2007; Kotagal & Broomall, 2012; Mazzone et al., 2018). Many of these conditions are worsened by poor sleep and thus may play a role in establishing and maintaining sleep disturbance. Further, medication prescribed to treat these issues (e.g., methylphenidate, risperidone, anti-epilepsy drugs) can accentuate sleep-interfering conditions such as pre-sleep arousal, restless legs, or daytime sleepiness (Cohen et al., 2014; Kotagal & Broomall, 2012; Mazzone et al., 2018; Trickett et al., 2018).

Parent perceptions/attributions. Recent research provides insight into sleep-related attributions held by parents of children with ASD. Beresford, Stuttard, Clarke, and Maddison (2016) found parents believed their child's diagnosis prevented them from effectively improving their child's sleep disturbance. Parents reported features such as, hyperactivity, increased anxiety, difficulty communicating, and hyper-responsivity to sensory stimuli, enhanced the complexity of implementing behavioural management strategies for sleep (Beresford et al., 2016). For example, parents left their child's bedroom lights on for reassurance and fulfilled food requests post-bedtime as they were unsure if their child was genuinely hungry (Kirkpatrick et al., 2019). Accordingly, some parents expressed a tendency to give in to child demands more easily as they had greater sympathy for their child due to their condition (Beresford et al., 2016). As a mother of a child with ASD, Farrar (2008) described lying with her daughter for hours when she engaged in vocalisations at bedtime. She dismissed comments advising her to lessen responses to her daughter's crying, as she believed her child was scared (Farrar, 2008). Similarly, other parents have reported treasuring the fact their child wished to fall asleep cuddling them, as opposed to falling asleep independently, despite this resulting in late nights (Dixon, 2006).

Kirkpatrick and colleagues' (2019) illustrated parents of children with ASD held varied perspectives regarding the value of a bedtime routine. While some parents considered a consistent bedtime routine to be of utmost importance given their child's need for structure,

order, and sameness, others felt establishing and maintaining a bedtime routine would be too difficult (Kirkpatrick et al., 2019). Additionally, although screen-based media is typically considered to be sleep-interfering, some parents believed it calmed their child and therefore enabled usage pre-bedtime (Kirkpatrick et al., 2019).

Technology. The increasing availability and popularity of screen-based technologies has resulted in increasing usage of this form of entertainment (Mazurek, Engelhardt, Hilgard, & Sohl, 2016). Many studies demonstrate exposure to television and video games before bedtime is associated with delayed sleep onset, reduced sleep duration, and daytime fatigue in typically developing children and adolescents (Mazurek et al., 2016). The mechanisms underlying this relationship are thought to relate to melatonin production, physiological or cognitive arousal, and content related psychological effects (Mazurek et al., 2016). Melatonin secretion at nighttime can be inhibited following exposure to bright light, thus interfering with the typical sleep-wake cycle (Mazurek et al., 2016). Exposure to electronic media, particularly violent video games, can heighten arousal levels, leading to difficulty initiating sleep (Mazurek et al., 2016). Lastly, viewing adult-themed content may result in anxiety or other unpleasant psychological states (Mazurek et al., 2016).

There is a distinct lack of research relating to technology use and sleep patterns among children with ASD (Mazurek et al., 2016). However, existing studies suggest children with ASD spend more time viewing screen-based media than typically developing children (Mazurek et al., 2016; Mazurek & Engelhardt, 2013; Mazurek & Wenstrup, 2013). In agreement with Kirkpatrick and colleagues' (2019) findings, Mazurek et al. (2016) suggest parents of children with developmental disorders may be more likely to use screen-based media to occupy and calm their children down, as well as to aid in behaviour management. Additionally, technology can become an obsessional or fixated interest for children with ASD (Chilvers, 2007). One parent of a 4-year-old with ASD commented "If I didn't stop her, she would watch TV all day with her face right up to the screen. If I turned it off, she would scream until it is switched on again" (Chilvers, 2007, p. 69).

Mazurek et al. (2016) conducted the first published study investigating the relationship between exposure to screen-based media before bedtime and sleep among children with ASD. The use of electronic media during the bedtime routine, as well as exposure to violent media content, was associated with significantly higher SOL compared to children whose bedtime routine did not include electronic media. Interestingly, this study found no significant difference

in SOL based on access to electronic devices in the child's bedroom. Conversely, Engelhardt, Mazurek, and Sohl (2013) found bedroom access to screen-based devices was associated with reduced sleep duration among typically developing children, and children with ASD or ADHD. Results of this study also indicated the effects of exposure to media were significantly worse for children with ASD (Engelhardt et al., 2013). As children with ASD are likely to have disrupted melatonin levels and experience difficulty regulating arousal already, the effects of technology on sleep may be markedly worse than for typically developing young people (Engelhardt et al., 2013).

Internal stimuli. For some individuals, inability to reach a state of behavioural quietude while in bed, due to intrusive cognitions, or an overactive mind, may create and maintain sleep difficulties (Didden et al., 2014). The prevalence of anxiety and depression is particularly high among young people with ASD and thus increases their risk of sleep disturbance (Lang, Regester, Lauderdale, Ashbaugh, & Haring, 2010; Nadeau et al., 2015; Richdale & Baglin, 2015). Autism-specific characteristics, such as cognitive inflexibility, alexithymia, poor emotional regulation, and communication difficulties, compound anxiety and depressive psychopathology present in typically developing young people (e.g., negative biases, unhelpful automatic thoughts, physiological arousal; Kotagal & Broomall, 2012; Ollendick & White, 2012). The presence of mood-related internal stimuli (e.g., racing thoughts, rumination) can prevent sleep onset when they occur close to bedtime (Mindell & Owens, 2015). Subsequent physiological (e.g., heart palpitations, stomach ache) and emotional distress (e.g., anxiety) can heighten unhelpful cognitive activity, producing a state antithetical to initiating and maintaining sleep (Harvey, 2005; Mazurek & Petroski, 2014). The relationship between sleep disturbance and internal stimuli is likely bidirectional (Uren et al., 2019). Cognitive and physiological hyperarousal can disrupt sleep initiation, and inadequate sleep further compromises self-regulation abilities, thus maintaining sleep-interfering internal stimuli (Nadeau et al., 2015).

Behavioural Theory of Sleep Disturbance

The behavioural model of sleep stems from research with typically developing young children (Blampied, 2013a; Blampied & France, 1993; France & Blampied, 1999; France, Blampied, & Henderson, 2003; France, Henderson**,** & Hudson, 1996), although it can be generalised to the whole lifespan (Blampied & Bootzin, 2013). In fact, the aetiology of sleep disturbance in young people with neurodevelopmental disorders is often behavioural (Beresford et al., 2016). Both classical and operant conditioning processes are involved in sleep and waking

and are therefore implicated in paediatric behavioural sleep medicine (Meltzer & McLaughlin Crabtree, 2015). While both processes are important to sleep, our understanding of the role of operant conditioning is much greater than that of classical conditioning (Blampied, 2013a; Blampied & Bootzin, 2013) and so the account below focuses on operant aspects.

Sleep itself is a biobehavioural state and not a behaviour, however, the transition into a sleep state does involve instrumental acts or operant behaviour (Blampied & Bootzin, 2013; Blampied & France, 1993). Operant behaviour is theorised to operate according to a three-term contingency involving the event or stimuli which precedes the behaviour (antecedent), the behaviour itself (response), and the subsequent state of the environment (consequence; Skinner, 1953). The process by which consequences strengthen the likelihood of a behaviour reoccurring is termed reinforcement (Blampied, 2013a). Positive reinforcement contingencies operate by adding a desirable stimulus to the environment as a consequence of a behaviour and negative reinforcement contingencies by removing an aversive stimulus from the environment in response to a behaviour (Blampied, 2013a). Critically, falling asleep can be both positively and negatively reinforcing, therefore, antecedents which precede the behaviour of falling asleep become discriminative stimuli for the occurrence of reinforcement (Blampied & Bootzin, 2013).

The state change from wake to sleep involves the completion of a behaviour chain, as the sleep-preparation to sleep initiation sequence is made up of numerous distinct units of behaviour (e.g., brush teeth, get into pyjamas, lie quietly in bed). Successful completion of each link in the behaviour chain exposes the individual to a stimulus event which is both a (secondary) reinforcer for the preceding behaviour and a discriminative stimulus for the behaviour of the next link and so forth (Blampied, 2013a). The chain ends when the individual enters the terminal link in which they emit the consummatory response which consumes the reinforcer of sleep (Blampied, 2013a). This consummatory response involves the maintenance of behavioural quietude for a sufficient period of time, accompanied by little or no cognitive and emotional arousal, in order for the early stages of sleep to occur (Blampied, 2013a; Blampied & France, 1993).

At any point during the bed-preparation and falling asleep behaviour chain an individual can choose to transition to other concurrent behaviour sequences/chains which lead to different reinforcers than the behaviour of going to sleep (Blampied, 2013a). The strength of secondary reinforcers in the bed-preparation and falling asleep behaviour chain are modulated by their respective distance away from the terminal reinforcer for the completion of the chain (sleep).

Therefore, the initial links in the bed-preparation and falling asleep behaviour chain are particularly vulnerable to disruption, as is the maintenance of behavioural quietude (Blampied & Bootzin, 2013), if the individual experiences discriminative stimuli for entering competing behaviour chains and competing reinforcers. Further, while sleep itself serves as the only reinforcer in the sleep initiation chain, multiple reinforcers may maintain sleep-interfering behaviours (Blampied & Bootzin, 2013). Additionally, competing behavioural repertoires are often highly salient, attractive, and immediately reinforcing (e.g., seeking parent attention, using electronic devices, engaging in stereotypy) and thus compared with the delayed reinforcement of sleep are likely to be chosen at any time over continuing with the bed-preparation chain (Blampied; 2013; Blampied & Bootzin, 2013).

It is therefore critical that the environment experienced by young people during their bed-preparation/falling asleep behaviour chain does not supply discriminative stimuli which control competitive behaviours, (e.g., electronic devices are not stored in the bedroom so device use is not an immediate option). Further, when modifying reinforcement contingencies which maintain sleep-conducive or sleep-interfering behaviour, it is critical to consider establishing operations. Establishing operations refer to antecedent events which increase (motivating operations) or decrease (abolishing operations) the reinforcement value of a behaviour (Michael, 1982). Given sleep deprivation is a core motivating operation for falling asleep and therefore increases the likelihood of behaviours which lead to this reinforcer (e.g., bed preparation; Michael, 1982) it is critical young people have sufficient physiological sleep pressure at bedtime. Establishing operations in this situation include ensuring young people do not make up for sleep loss at night by sleeping in or napping during the day and going to bed when they are sleepy. In addition, increasing the availability of reinforcers for sleep-interfering behaviour prior to bed (e.g., social attention, access to tangibles) satiates the value of these as reinforcers post-bedtime and reduces the likelihood they will occur (Jin et al., 2013).

Parent behaviour is often a component of the bed-preparation/falling asleep behaviour chain as parents tend to instruct and supervise their child's bedtime routine (Blampied & Bootzin, 2013; Blampied & France, 1993). Consequently, it is often the parent's role to modify establishing operations and reinforcement contingencies for sleep-interfering (e.g., minimise response to disruptive behaviour post-bedtime) and sleep-conducive behaviour (e.g., provide descriptive praise). Parents can support their child to enter and maintain behavioural, emotional, and cognitive quietude through a calm, quiet bedtime routine (Blampied, 2013a).

When falling asleep is under strong stimulus control (i.e., discriminative stimuli in the behaviour chain process have been consistently associated with sleep onset and reinforced by sleep itself), sleep onset is likely to occur in the presence of these discriminative stimuli (Blampied, 2013a). Notably however, a range of interoceptive (e.g., tiredness), proprioceptive (e.g., posture), and exteroceptive (e.g., nighttime) discriminative stimuli may be necessary to initiate sleep (Blampied, 2013a; Blampied & Bootzin, 2013). For example, an individual is unlikely to fall asleep in the presence of external discriminative stimuli alone without physiological pressure to fall asleep. The absence of consistent or appropriate discriminative stimuli which signal the initiation of bed-preparation and falling asleep behaviour chains may contribute to sleep disturbance (Blampied, 2013a; Blampied & France, 1993). Inappropriate discriminative stimuli for sleep may include those which a young person cannot produce themselves (e.g., cuddling parent), are sleep-interfering (e.g., using a handheld electronic device), or involve a location other than their typical sleep environment (e.g., parent's bed). Critically, if discriminative stimuli for sleep are not available throughout the night the individual is likely to have difficulty re-establishing sleep upon waking, instead requiring the reinstatement of the stimulus conditions present at initial sleep onset (Blampied, 2013a).

Parents and their children can fall into a coercive behaviour trap (Patterson, 1982) whereby inappropriate discriminative stimuli for sleep and sleep-interfering behaviours are maintained through a double reinforcement contingency (Blampied & France, 1993). This is double in that both the child and parents act to initiate established sleep behaviour chains to avoid aversive circumstances (France et al., 2003). For example, a child who has learned that the discriminative stimuli for falling asleep are those provided by their parents' presence, is likely to engage in sleep-interfering behaviour (e.g., calling out) if this requirement is not met (France et al., 2003). This resistance may reflect the child's effort to avoid the potential anxiety of falling asleep independently (France et al., 2003). The parent may reinitiate the inappropriate discriminative stimuli for sleep to minimise the child's distress and establish sleep in the short-term (France et al., 2003). The reinforcement contingency is also double in that it involves both negative and positive reinforcement. Both parties are negatively reinforced through avoidance of distress and positively reinforced through initiation of sleep in the short-term. To avoid distress and initiate sleep, both parent and child are likely to continue engaging in this same behaviour pattern in anticipatory ways (France et al., 1996). Furthermore, these same settling events are likely to be required if the child wakes during the night in order to re-establish sleep (France et al., 2003).

The likelihood of falling into a sleep-related behaviour trap is influenced by a multitude of factors, including both parent and child characteristics and temperament, as well as parenting practices, parental self-efficacy, and mental health (France & Blampied, 1999). Aspects such as, stress, marital discord, mental health problems, and anxiety are more likely to be intensified in parents of children with developmental disabilities (Richdale & Wiggs, 2005). Consequently, they may find it more difficult to maintain consistent, disciplined approaches to their child's bedtime routine and encourage independent sleeping practices (Richdale & Wiggs, 2005). Further, children's own sleep-related anxiety, such as fear of the dark or separation from parents, also contribute to sleep-interfering behavioural chains.

Although operant conditioning procedures have been emphasised thus far, classical conditioning processes are also implicated in the development and maintenance of sleep disturbance. In fact, insomnia may develop and persist due to chronic, conditioned arousal (Blampied & Bootzin, 2013). Individuals with insomnia typically spend extended periods of time in bed awake, feeling frustrated or anxious regarding their lack of sleep (Bootzin, Smith, Franzen, & Shapiro, 2010). Consequently, their typical bedtime environment can become a conditioned stimulus for conditioned responses of physiological hyperarousal, instead of sleep (Bootzin & Epstein, 2011; Mindell & Owens, 2015). A similar process may occur with young people who are frightened by aspects of their sleep setting (e.g., darkness, being alone) and have developed a conditioned fear response to this environment (Blampied & Bootzin, 2013).

Notably, conditioned stimuli (e.g., the sleep environment) for arousal or arousal inhibition may also function as discriminative stimuli for sleep-interfering (e.g., electronic device use) or sleep-facilitative (e.g., lying quietly) behaviour at the same time. For example, when individuals with insomnia engage in sleep-interfering behaviours while in bed (e.g., reading, using screen-based media), the bedroom environment may become both a conditioned stimulus for wakefulness and a discriminative stimulus for sleep-interfering behaviour (Bootzin et al., 2010; Bootzin & Epstein, 2011). Consequently, appropriate and available stimuli, such as the bed or bedroom no longer serve as conditioned stimuli for arousal inhibition or discriminative stimuli for sleep (Bootzin & Epstein, 2011). Ideally, sleep-facilitative conditioned stimuli that produce the conditioned response of relaxation, should be congruent with discriminative stimuli which signal the initiation of falling asleep behaviour chains, ending in the terminal link and the consummatory response of sleep. If they are incongruent, sleep onset difficulties are likely to occur.

During intervention elements of the sleep behaviour chain need to be brought under appropriate stimulus control, and contingencies of reinforcement should strengthen and maintain this behaviour chain as well as other sleep compatible behaviours (Blampied & France, 1993). Additionally, any inappropriate conditioned stimuli (i.e., those that induce physiological arousal) need to be addressed via appropriate strategies, such as habituation or desensitization (Blampied, 2013a). Effective intervention is more likely if stimuli which interfere with behavioural quietude and prevent sleep onset are recognised (Jin et al., 2013). Behavioural principles, notably functional behavioural assessment (FBA: Blampied, 2013a) are necessary to identify many common contributing factors to young people's sleep disturbance and inform effective treatment approaches for each family. The complex chain of events which reinforce and maintain sleep disturbance are unique to each individual.

FBA

FBA is a method used to identify the relationship between an antecedent, behaviour, and its consequence/s in order to discover the purpose or function the behaviour serves and to identify the nature and role of any relevant conditioned stimuli affecting the target behaviour (Blampied, 2013a). FBA evolved from the literature regarding experimental functional analysis, which was developed by Iwata and colleagues' in 1982. Their seminal research showed that positive, negative, and automatic reinforcement contingencies underlying problem behaviour can serve four functions: attention (e.g., social interactions), tangibles (e.g., electronic device); escape (e.g., avoidance of perceived aversive event), and sensory stimulation (behaviour maintained by sensory consequences, such as stereotypy; Iwata, Dorsey, Slifer, Bauman, & Richman, 1994; Iwata &Worsdell, 2005). They demonstrated how directly observing problem behaviour in real-time, while systematically manipulating reinforcement contingencies, can determine which of these functions underlie challenging behaviour. This procedure was coined functional behaviour analysis. Since this time, numerous empirical studies have consistently demonstrated behavioural interventions preceded by functional analysis are significantly more effective than standard behavioural interventions not preceded by this assessment method (e.g., a manualised approach; Campbell, 2003; Didden, Korzilius, van Oorsouw, & Sturmey, 2006; Harvey, Boer, Meyer, & Evans, 2009; Heyvaert, Saenen, Campbell, Maes, & Onghena, 2014; Scotti, Evans, Meyer, & Walker, 1991).

While functional analysis can verify behaviour function/s when they are ambiguous (Iwata & Worsdell, 2005), this standardized experimental approach has significant practical

(e.g., time-intensive for family members and clinician, implementation throughout the night) and ethical barriers (e.g., purposefully providing reinforcement for problem behaviour) when applied in a sleep context (Blampied & Bootzin, 2013). Accordingly, experimental functional analysis is rarely used in the development of paediatric sleep interventions (Blampied, 2013a). In contrast to functional analysis, FBA involves indirect assessment methods (e.g., clinical interviews, questionnaires), direct observation (e.g., videosomnography), and/or records of direct observation (e.g., behaviour diary) of problem behaviour in its natural environmental context, without any form of manipulation (Blampied & Bootzin, 2013).

FBA is useful within a sleep context to identify the relationship between external environmental or internal (private) events and ongoing sleep problems, ensuring appropriate modification of antecedent contexts, actual and potential reinforcers, and interfering and facilitative conditioned stimuli (Blampied, 2013a; Kodak & Piazza, 2008). Lack of correspondence between behaviour and its function may compromise intervention success (Kodak & Piazza, 2008). In fact, failures in the treatment of sleep disturbance may be due to targeting behavioural topographies that appear similar across cases, as opposed to addressing the function of each individual's sleep-interfering behaviour (Brown & Piazza, 1999; Kodak & Piazza, 2008). For example, leaving the bedroom post-bedtime may be functionally maintained by social attention for one young person, and access to tangibles for another, each indicating different interventions. Critically, Iwata and colleagues (1994) showed a single behavioural topography can serve multiple functions. For example, engagement in stereotypy may be reinforced by social and automatic reinforcement, thus the function of the behaviour is to seek both sensory stimulation and parent attention.

In accordance with functional analysis, FBA is also a rigorous and empirical technique which maximises the efficiency and effectiveness of behavioural interventions by identifying the unique contributions to problem behaviour and thus informing targeted, individualised treatment (Myers & Plauché Johnson, 2007; Roane, Fisher, & Carr, 2016). Although functional assessment processes are commonly used within the assessment and treatment of general behaviour problems in young people with ASD, they have been applied to sleep disturbance relatively rarely (Jin et al., 2013).

Including Young People in Research

Following the introduction of the United Nations (UN) Convention on the Rights of the Child (1989) the importance of including children with a disability in research relating to their

lives has been widely recognised (Harrington, Foster, Rodger, & Ashburner, 2013). Both national and international human rights legislation and codes of ethics recognise the importance of inclusive practices when engaging with young people with disabilities. According to the *Convention on the Rights of Persons with Disabilities* (UN, 2006), people with disabilities should be granted individual autonomy, including the ability to make their own choices; have equality of opportunity; and experience full and effective inclusion and participation within society. Within Article 7, the UN convention declares children with disabilities shall be able to express their views freely on topics which affect them (UN, 2006). The *Code of Ethics for Psychologists working in Aotearoa/New Zealand* (New Zealand Psychological Society, 2012), also states that psychologists are obligated to ensure full and active participation from individuals in relation to decisions which impact them. These documents reflect the basic human right for all individuals to be included to the maximum degree possible in all issues concerning them. This thesis aimed to show how it is possible to act in accordance with these guidelines by promoting the participation and active involvement of young people with developmental disabilities in understanding and treating their sleep disturbance.

Young people value the opportunity to share their opinions (O'Kane, 2008; Roberts, 2008). Children with disabilities report they would like to be involved in decisions regarding how they spend their time, future plans, and for professionals to talk with them and provide them agency (Beresford, Rabiee, & Sloper, 2007); however, young people with disabilities are more commonly excluded from research than those without disabilities (Bailey, Boddy, Briscoe & Morris, 2015). While parents are often included as active agents within research, young people tend to be involved as passive agents (Christensen & James, 2008). This dismisses the importance of their own experiences, perspectives, and knowledge to inform future research and clinical practice (Woodhead & Faulkner, 2008). Conventional research tends to explore young people's worlds through the perspectives and understanding of their adult caregivers (Christensen & James, 2008). Often adult knowledge is viewed as superior to young people's knowledge (Mayall, 2008). However, only young people with ASD experiencing sleep disturbance have knowledge about their own unique experience and what this feels like to them.

Involving young people with disabilities is critical to uncovering what they feel works for them and their families (Bailey et al., 2015). However, the perspectives of young people with ASD (e.g., views regarding treatment acceptability) have been rarely sought within research (Harrington et al., 2013). Commonly, young people are excluded from research due to concerns regarding their cognitive and language abilities and subsequent capacity to discuss their

behaviour, perceptions, and beliefs (Scott, 2008). Harrington and colleagues (2013) suggest diagnosis-related assumptions may prevent the direct involvement of young people with ASD in research. However, the heterogeneity of individuals on the autism spectrum means homogenous assumptions about impairment should not inform decision making regarding their involvement. Further, individualised adaptations can be made to facilitate the involvement of young people with disabilities. Bailey et al. (2015) conducted a systematic review of all studies published since 1990 that involved children with disabilities as active agents in research. Results suggest appropriate modifications include regular breaks, scheduling sufficient time, use of engaging and interesting activities, providing the child with a choice of activities if possible, and utilising a flexible approach to allow for diverse abilities (Bailey et al., 2015). Research shows young people with ASD are able to provide insight into their world with appropriate support (Harrington et al., 2013).

Involving young people with disabilities in research has been shown to result in improved research quality; increased confidence, self-esteem, and independence; acquisition of new skills and experiences; empowerment; feeling their perspective is valued; and fulfilment in the knowledge they may help other young people (Bailey et al., 2015). Nevertheless, involving young people with developmental disabilities in research does not occur without risk. Involvement can lead to feelings of intimidation when collaborating with unfamiliar adults in unknown environments and young people may feel less confident in their abilities when faced with seemingly impossible tasks (Bailey et al., 2015). Roberts (2008) notes time is one of the few resources young people have, therefore, researchers need to ensure the appropriateness of asking them to donate this. Further, adaptations to enable their involvement is a time-consuming process, requiring more resources, training, skills, and extensive preparation (Bailey et al., 2015; Beresford, Tozer, Rabiee, & Sloper, 2004; Harrington et al., 2013).

Another important and complex consideration when involving young people with or without disabilities in research, is its salience to them. Due to developmental factors, caregivers and/or professionals often have the right to decide what is in the best interests of young people, however, this may be in direct contrast to the wishes of children and adolescents (France, Annan, Tarren-Sweeney, & Whitcombe-Dobbs, 2016; Kendall, 2018; Weisz & Jensen, 1999). As mentioned previously, sleep-interfering behaviour (e.g., device use in bed) is often highly reinforcing for young people, despite potentially contributing to poor long-term outcomes. Young people may not be motivated to participate in research which relates to their engagement in maladaptive behaviour (Salari, Ralph, & Sanders, 2014). Further, as with any participant,

young people's input may need to be considered in light of potential predispositions, such as perceiving sleep-interfering behaviour favourably.

Conclusion

Although the appropriateness of actively involving young people with developmental disabilities within research and therapeutic processes needs to be carefully considered, national and international codes and legislation emphasise that the presence of a developmental disability should not preclude their involvement. While parents and caregivers play a critical role in the development, implementation, and evaluation of sleep interventions, so too can young people themselves.

Sleep disturbance is a significant issue for many young people with ASD and can be detrimental to their own and their family's wellbeing. As discussed above, a number of factors may be implicated in the establishment and maintenance of sleep disturbance in young people with ASD. Behavioural models of sleep disturbance are largely based on typically developing young children, although when generalised to ASD-related sleep problems they can reveal modifiable contributing factors. Utilisation of FBA may be critical to inform individualised, targeted, and effective sleep interventions for young people with ASD. The effectiveness of FBA-informed behavioural sleep interventions which include both parents and young people with ASD in the assessment, treatment, and evaluation process are in need of investigation.

Chapter 2

Literature Review

Interest in the sleep of young people with ASD has risen substantially in recent years (Beebe, 2016; Meltzer, 2016). More publications relating to sleep in young people with ASD have been published since 2010 than were published in all indexed years prior to this (Beebe, 2016). Most research has focused on whether sleep disturbance is comorbid with some other condition, exacerbates symptomatic behaviour, or is the result of the disorder itself (Beebe, 2016). Less research has explored the utility of behavioural sleep interventions for young people with ASD. The following section includes a review of published studies implementing behavioural sleep interventions with children and adolescents with ASD, and the use of FBA to inform sleep treatment. In accordance with a human rights and ethical approach, the inclusion of young people as active agents within the preceding research is explored. Additionally, the effectiveness of interventions which include young people with ASD in the therapeutic process is evaluated.

The purpose of this literature review is to establish the current evidence for behavioural sleep interventions to treat children and adolescents with ASD and explore the extent to which FBA has been used within this area. As the aim of this thesis is to actively include young people with ASD in the therapeutic process, it is necessary to establish the degree to which this has been done in previous research. Research regarding sleep interventions applied to typically developing young people is explored to ascertain how children and adolescents can be included as active intervention agents. Due to differences in neurological functioning, appropriate modifications which facilitate the inclusion of young people with ASD in the therapeutic process need to be identified. Finally, a review of interventions which include young people with ASD as active agents is necessary to determine the evidence base for these inclusionary methods. Findings were used to inform assessment and intervention practices within the current research.

Behavioural and Cognitive Sleep Interventions Applied to Autism-related Sleep Disturbance

Search process. In order to find relevant literature regarding sleep interventions for young people with ASD, databases searched included PsycInfo; Psychology and Behavioral Sciences Collection; ERIC; and Embase. The searches combined the terms: “autism spectrum disorder”, “ASD”, “Asperger syndrome”, “pervasive developmental disorder”,

"neurodevelopmental disorder", "sleep*", "insomnia", "dyssomnia", "parasomnia", "sleep onset latency", "sleep interfering behavio*r", "night waking", "night awakening", "bedtime resistance", "bedtime refusal", "curtain calls", "intervention", "treatment", "therapy", "functional behavio*r assessment", and "cognitive behavio*r therapy". Pearling was conducted to discover articles not located by the database searches. Studies were included in the review if they met the following criteria: (a) participants were 2 to 18 years old and had a diagnosis of autism, Asperger's, PDD, or PDD-NOS (if the study included other diagnostic categories [e.g., children with ADHD], results for young people on the autism spectrum had to be reported separately); (b) participants experienced sleep disturbance (e.g., bedtime resistance, frequent and extended NWs); (c) sleep was systematically measured via one or more of the following measures: polysomnography (PSG; records physiological activity, such as brainwaves, breathing, and eye movements to assess sleep architecture and is conducted overnight in a laboratory), videosomnography (VSG; continuous infrared video recording of the young person in their natural sleep environment throughout the night), actigraphy (wrist worn device which uses limb movement as a proxy measure for sleep), parent- and/or self-report sleep diaries (daily sleep logs) or questionnaires (e.g., the Children's Sleep Habits Questionnaire, [CSHQ; Owens, Spirito, McGuinn, 2000] a parent-report questionnaire for assessing school-aged children's sleep patterns); (d) a behavioural or cognitive behavioural sleep intervention was conducted; and (e) the article was written in English. The search yielded 13 intervention approaches, including consistent bedtime routine, sleep hygiene, unmodified extinction, graduated extinction, systematic fading of parental presence, social stories, bedtime fading with or without response-cost, sleep restriction, scheduled awakening, chronotherapy, parent education programmes, young person education programmes, and cognitive behavioural therapy (CBT).

Consistent bedtime routine/ sleep hygiene. Behavioural approaches were first implemented in the treatment of sleep disturbance in children with ASD in the 1960s (Schreck, 2001). Wolf, Risley, and Mees (1963) were the first known researchers to apply operant conditioning principles in the treatment of sleep disturbance in a child with ASD. A consistent bedtime routine combined with unmodified extinction including mild punishment (the child's bedroom door was closed if he left his room or engaged in tantrums during NWs) was able to reduce the 3.5-year-old participant's NW. A consistent bedtime routine is thought to establish discriminative stimuli for sleep onset (ensuring sleep is under strong stimulus control) and can help regulate natural melatonin levels and one's circadian rhythm (Katz & Malow, 2014;

Shreck, 2001). A consistent bedtime routine can also facilitate sleep in children with ASD who insist on sameness (Katz & Malow, 2014). While a number of studies within the ASD and sleep literature have recommended that families maintain consistent sleep-conducive bedtime routines, this treatment component has only been evaluated independently within one study. Delemere and Dounavi (2018) implemented a consistent bedtime routine with three children with ASD aged 2 to 6 years. Parents were required to construct an appropriate routine with therapist support and introduce this to their child using a visual schedule. Applied independently of other intervention strategies, this technique did not significantly reduce sleep disturbance (Delemere & Dounavi, 2018). Despite this, parent ratings of treatment acceptability were high.

The term sleep hygiene refers to habits which promote appropriately timed and effective sleep (Hauri, 1977; Jan et al., 2008). In addition to maintaining a bedtime routine, sleep hygiene may include avoiding stimulating activities before bedtime and ensuring optimal sleep conditions, such as a dark room, adequate temperature, and low external noise (Singh & Zimmerman, 2015; Vriend et al., 2011). Good sleep hygiene also involves minimising the following daytime behaviours: naps, exercise within 60 minutes of bedtime, and caffeine intake (Singh & Zimmerman, 2015). While sleep hygiene alone may not be sufficient to treat sleep disturbance, education regarding the importance of good sleep hygiene is a necessary component of sleep interventions (Kodak & Piazza, 2008; Vriend et al., 2011) and is often the first line of treatment (Jan et al., 2008). Sleep hygiene as well as behavioural therapy have demonstrated effectiveness for both typically developing children and children with ASD (Cortesi et al., 2010).

Extinction. Extinction procedures are applied when sleep-interfering behaviour is reinforced by access to attention, tangibles (including eating and drinking), or escape from an undesired event, such as being put to bed (Didden et al., 2014). Extinction involves eliminating reinforcement for sleep-interfering behaviour to reduce or extinguish such behaviour (Didden et al., 2014). In the sleep literature, extinction procedures which do not involve any procedural modifications to ameliorate side-effects (e.g., emotional distress) are referred to as ‘unmodified extinction’ or ‘unmodified planned ignoring’. Unmodified extinction of socially maintained sleep-interfering behaviour would require parents not to attend to any bedtime disruptions, such as crying or calling out (Vriend et al., 2011). If the child leaves their bedroom, parents are instructed to return the child to their bed with minimal engagement. Unmodified extinction procedures are initiated all at once on a designated night and maintained thereafter. As mentioned previously, Wolf et al. (1963), implemented unmodified extinction at an inpatient

ward with a young child with ASD who was experiencing delayed sleep onset and night wakings. Each time the child left his room he was returned by staff without engagement and his bedroom door was closed. Although this intervention was successful, during the first five nights of treatment the child engaged in extended and violent tantrums. This increase in challenging behaviour following implementation of extinction strategies is the phenomenon termed a 'post extinction response burst' (France et al., 1996) or 'extinction burst' in the applied behaviour analysis literature (Lerman, Iwata, & Wallace, 1999). PERBs are a well-documented feature of extinction in both basic and applied research (Lerman & Iwata, 1995) and are commonly observed in the initial phase of extinction. They are paradoxical, in that they involve an initial increase in the frequency, intensity, and/or duration of a behaviour that is no longer receiving reinforcement. This usually occurs prior to a reduction in target behaviour (France et al., 1996). In applied research, PERBs are problematic as children's crying and protests can be distressing to parents, making it difficult to minimise their typical responses (Kodak & Piazza, 2008). The risk of an unmodified extinction procedure is that parents may eventually give in and respond to their child at the peak of the child's disruptive behaviour (Kodak & Piazza, 2008). This is likely to result in the child escalating their disruptive behaviour further during the following nights as escalation has previously been successful in gaining reinforcement (Kodak & Piazza, 2008). When using unmodified extinction, it is critical parents receive regular support and guidance to maintain treatment fidelity and ensure effective implementation (Vriend et al., 2011), this includes being warned about and prepared to cope with PERBs if they occur (France et al., 1996).

Four studies were identified which successfully implemented unmodified extinction in children aged 3 to 7 years with Asperger's and ASD (Didden, Curfs, van Driel, & de Moor, 2002; Weiskop, Matthews, & Richdale, 2001; Weiskop, Richdale, & Matthews, 2005; Wolf et al., 1963). Of note, the procedures implemented by Wolf et al. (1963) were conducted within an inpatient ward and thus the results may differ when delivered within home-based settings as in this thesis. Weiskop et al. (2001; 2005) supplemented unmodified extinction with reward systems for engagement in sleep-conducive behaviour and maintenance of bedtime routines. Parent education was also a main component of the intervention (Weiskop et al., 2001; 2005). During four training sessions within the participant's home, therapists educated parents in behavioural theory, instructed them in the use of reinforcement procedures and extinction techniques, and taught them how to provide effective instructions (Weiskop et al., 2001; 2005).

Treatment improved self-regulation at night, unwanted co-sleeping, NWs, and disruptive behaviour (Weiskop et al., 2001; 2005).

More recently a few studies have implemented unmodified extinction within FBA-informed multicomponent sleep interventions. Jin et al. (2013), McLay, France, Blampied, Danna, and Hunter (2017), McLay, France, Blampied, and Hunter (2019), and McLay, France, Knight, Blampied, and Hastie (2019) restricted access to putative reinforcers (e.g., books, social attention) post-bedtime for the remainder of the night to reduce sleep-interfering behaviour. Alongside additional behavioural components (e.g., positive reinforcement for sleep-conducive behaviour), these interventions were successful in reducing sleep-interfering behaviour and associated sleep disturbance. Further, treatments were generally rated as acceptable by parents (McLay, France, Knight et al., 2019).

Implementation of unmodified extinction and bedtime routines has been effective in improving settling and reducing co-sleeping and NWs in children with developmental disabilities with diverse functioning (Richdale & Wiggs, 2005). According to Chambless and Hollon's (1998) treatment efficacy criteria, unmodified extinction in children with ASD meets the possibly efficacious standard, having proved beneficial to at least three participants within a methodologically sound study (Vriend et al., 2011). Independent replication of these studies is required before the use of unmodified extinction alone can be considered a well-established sleep intervention for young people with ASD.

Graduated extinction. Unmodified extinction is not always feasible or desirable. Firstly, parents may be unwilling to carry out this procedure with complete fidelity, and secondly, it is not appropriate if children with ASD are likely to engage in destructive or self-injurious behaviour when distressed or unattended (Ferber, 1985; Kodak & Piazza, 2008). Alternative treatment methods include graduated extinction and systematic fading of parental presence. Application of graduated extinction allows reinforcement for sleep-interfering stimuli to be reduced progressively, as opposed to immediately according to unmodified extinction procedures. For example, in response to sleep-interfering behaviour caregivers could provide minimal reassurance non-contingently according to a predetermined schedule (e.g., every 5 minutes; Turner & Johnson, 2013). The interval between checking can then be gradually increased over time (Turner & Johnson, 2013). This treatment method allows parents to continue checking their child while gradually reducing the attention received. As parental

checking is non-contingent on the child's disruptive behaviour, it is anticipated to be less likely to be reinforced through social attention.

Durand, Gernet-Dott, and Mapstone (1996) implemented graduated extinction in conjunction with sleep hygiene with two children on the autism spectrum (one with PDD). The graduated extinction procedure consisted of systematically increasing the amount of time before their caregiver responded to bids for attention post-bedtime over subsequent nights (e.g., 3 min first night, 5 min second night). Following intervention, both children experienced a reduction in disruptive behaviour (e.g., tantrums) post-bedtime and one demonstrated improved SOL. Graduated extinction was implemented alongside a social story (described later) within a study by Moore (2004) to treat bedtime resistance, co-sleeping, delayed SOL, NW, and EW in a 4-year-old boy with ASD and learning disabilities. Following treatment, disruptive behaviour at bedtime was reduced, co-sleeping eliminated, and SOL improved. Further, the participant's mother perceived the intervention to be successful and easy to implement. Knight and Johnson (2014) implemented graduated extinction alongside a developmentally appropriate bedtime, consistent sleep routine, and white noise in three young children with ASD. This multicomponent intervention successfully reduced SOL and NWs, however the contribution of each individual component to the outcome is unknown. Other graduated extinction interventions have been carried out with children with ASD through a parent training booklet (Montgomery, Stores, & Wiggs, 2004) and group family workshops (Malow et al., 2014; Reed et al., 2009). Most children within these studies showed some improvement in sleep disturbance. More research evaluating graduated extinction use with children with ASD is necessary before the efficacy of this technique can be established (Turner & Johnson, 2013; Vriend et al., 2011).

Systematic fading of parental presence. To date, fading of parental presence has been employed in four published studies including children with ASD, to address unwanted co-sleeping (Howlin, 1984; McLay et al., 2017; McLay, France, Knight et al., 2019; Souders et al., 2017). In Howlin's (1984) study, the parent of a 5.5-year-old boy with ASD slept on a mattress next to his bed, with the mattress systematically moved further away over an 8-week period. Following this intervention, SOL improved and co-sleeping was eliminated, however, NW still occurred approximately two or three times a week. Additional procedures in conjunction with stimulus fading may be necessary to resolve a range of sleep problems. Other studies have successfully faded parent presence by initially instructing parents to sit or lie beside the child's bed during sleep onset and NWs, then systematically decreasing this proximity over a number of

weeks until they are out of sight (McLay et al., 2017; McLay, France, Knight et al., 2019; Souders et al., 2017).

Social stories. As briefly mentioned earlier, social stories can also be used in the treatment of sleep disturbance in young people with ASD. Although, initially intended to teach social skills, social stories are now used as tools to educate individuals with ASD about a range of appropriate behavioural sequences (e.g., toilet training; Gray, 2010). Traditionally, social stories are presented in a storybook format containing short, positively framed sentences, written in first- or third-person, with corresponding pictures. They may be generic, with applicability to many children and situations, but most often they are individualised to a specific child or children and/or contexts and situations. Their rationale is drawn from Bandura's Social Learning Theory (Schneider & Goldstein, 2010) with the character in the story acting as a model for some prosocial behaviour. Social stories have been used effectively to teach children with ASD socially appropriate behaviours and skills, but no published studies were found using social stories as the primary treatment for sleep disturbance in young people with ASD.

Nonetheless, an unpublished study indicates the importance of further research in this area. Social stories were included as a core element within a sleep intervention for 11 children (3 to 9 years) with ASD as part of a doctoral thesis (Gilles, 2008). Alongside parent psychoeducation and sleep hygiene (phase one), a picture schedule and social story was created for each family (phase two). Parent-child dyads attended a 1-hour session with a clinician whereby they read through the social story together, and then problem solved how to use the social story and picture schedule to reduce sleep disturbance. Phase two resulted in reduced SOL, bedtime resistance, and NWs for all participants. However, it is difficult to determine the exact impact of this phase due to ordering effects. Nevertheless, parents reported their children often requested the social story at bedtime and enjoyed this intervention method. Gilles (2008) suggests the story may have increased the frequency of positive parent-child interactions at bedtime, thus reinforcing children's compliance and potentially creating an appropriate discriminative stimulus for sleep.

Other studies have incorporated social stories in multicomponent behavioural sleep interventions to introduce and explain new sleep routines and reinforcement systems to young people with ASD (McLay et al., 2017; McLay, France, Knight et al., 2019; McLay, France, Blampied et al., 2019; Moore, 2004; Souders et al., 2017). As mentioned previously Moore (2004) developed a social story illustrating the child's bedtime routine and consequences. The

social story was intended to help the child understand the behavioural programme as well as remind parents of the steps they must follow each night in order to remain consistent (Moore, 2004). To increase the child's engagement when reading the story, pictures of items in his bedroom were cut out with Velcro fixtures so he could arrange the pictures appropriately each time the story was read. Additionally, pictures of well-liked characters including 'Thomas the Tank Engine' and 'Tigger' were used in the story. Additional treatment strategies included a reward system whereby the child fastened Velcro 'Thomas the Tank Engine' pictures to a chart each night he engaged in sleep-conducive behaviour. Before bedtime the child was also required to place a 'do not disturb' sign on his parent's door and a 'Thomas' door hanger on his own bedroom door. These techniques reduced disruptive behaviour at bedtime, eliminated co-sleeping, and improved SOL.

Similarly, McLay et al. (2017), McLay, France, Blampied et al. (2019), McLay, France, Knight et al. (2019), and Souders et al. (2017) have used social stories as part of multicomponent treatment packages effective in eliminating co-sleeping and reducing SOL and duration of NWs. As social stories have typically been used in conjunction with other methods, the effectiveness of social stories in isolation is unclear. Nevertheless, social stories may provide a valuable addition to treatment and can be useful for children who have significant communication difficulties (Richdale & Wiggs, 2005).

Bedtime fading with or without response-cost. Bedtime fading consists of delaying bedtime to increase homeostatic sleep pressure and the likelihood of rapid sleep initiation. It reduces time spent in bed awake (ensuring the bedroom environment becomes a discriminative stimulus for sleep) and increases the reinforcement value of sleep onset. Bedtime fading was derived from Bootzin's (1977) Stimulus Control Therapy for Insomnia in adults. The process involves delaying a young person's bedtime to within 15 minutes of their typical sleep onset time, eliminating their daytime sleep, and establishing a set wake time (Piazza & Fisher, 1991a; 1991b). The bedtime can be moved earlier in small increments when they can fall asleep quickly with little resistance (Kodak & Piazza, 2008). This procedure continues until the desired bedtime is reached (Kodak & Piazza, 2008). Bedtime fading with response-cost follows the same basic method, however if the young person does not fall asleep within 15 minutes, to reduce time in bed spent awake, they are removed from their bed to engage in a quiet, non-stimulating activity for a short time period (Vriend et al., 2011). This is repeated until they fall asleep (Vriend et al., 2011).

Six studies have implemented bedtime fading without response-cost in the home setting (Delemere & Dounavi, 2018; Jin et al., 2013; McLay, France, Blampied et al., 2019; McLay, France, Knight et al., 2019; Moon et al., 2011; Sanberg, Kuhn, & Kennedy, 2018) and one in an inpatient setting (DeLeon, Fisher, & Marhefka, 2004). Moon and colleagues (2011) provided a treatment handbook to parents of three children (8 to 9 years of age) with ASD, which described implementation of bedtime fading and a reward programme. Long-term improvements in SOL were evident at 3-month follow-up. However, there was no clear change in participant sleep duration and SE post-treatment. Additional results indicated a small improvement in the children's daytime behaviour, and parent treatment acceptability fell within the average range. Jin et al. (2013), McLay, France, Blampied et al. (2019), and McLay, France, Knight et al. (2019) incorporated bedtime fading without response-cost within FBA-informed multicomponent treatment programmes for children aged 2 to 10 years. Although the interventions were relatively effective, the contribution of bedtime fading is unclear as components were not implemented independently of one another.

Sanberg et al. (2018) and Delemere and Dounavi (2018) investigated the effectiveness of bedtime fading without response-cost alone. Across both studies this treatment strategy resulted in reduced SOL and increased total sleep duration amongst children aged 2 to 6 years. Interestingly, Sanberg et al. (2018) found bedtime fading without response-cost was effective in eliminating unwanted co-sleeping during the night and at sleep onset also. Further, treatment resulted in a reduction in bedtime resistance and NWs. Parents rated bedtime fading as highly acceptable.

Bedtime fading with a response-cost procedure has only been applied in an inpatient setting with 6- to 8-year-old children with ASD (Piazza, Fisher, & Sherer, 1997). While treatment eliminated EWs for one participant, the remaining two participants continued to experience difficulty initiating and maintaining sleep (Piazza et al., 1997). If participants did not fall asleep within 15 minutes, they were kept awake for 1 hour during which they were allowed to engage in the activity of their choice, play with toys, or watch television. Such activities may be too stimulating for some children and thus inhibit their ability to return to sleep post response-cost. Additionally, the severity of participant behaviour within this study, as well as the pro-longed monitoring necessary to implement bedtime fading with response-cost, may be difficult to carry out within a home setting where access to supervised care throughout the night would not be feasible.

Overall, there is increasing evidence for bedtime fading without response-cost for young children with ASD and it is a well-established intervention for children with an ID (Didden et al., 2014), however, the efficacy of this intervention for older children and adolescents with ASD has not yet been established.

Sleep restriction. Sleep restriction is a similar approach to bedtime fading and also has its origins in the treatment of adult insomnia. When implementing sleep restriction, the amount of time a young person spends in bed is limited to 90% of the total amount of time they typically sleep (Vriend et al., 2011). Equivalent to bedtime fading with response-cost, if the child does not fall asleep within 15 minutes they are removed from their bed and engaged in a non-stimulating activity (Vriend et al., 2011). If the child falls asleep within a short time frame, their bedtime is gradually brought forward until the desired time is reached (Vriend et al., 2011). Durand and Christodulu (2004) implemented sleep restriction with a 4-year-old girl with ASD demonstrating severe tantrums at bedtime and during NWs. The child's bedtime was delayed to midnight while her 7:00am wake up time remained consistent (Durand & Christodulu, 2004). Following intervention, the child's disruptive behaviour at bedtime reduced, as well as the frequency of NWs and disruptive behaviour during these awakenings. She was also able to cease melatonin usage (Durand & Christodulu, 2004). But, concerningly, she experienced an increase in sleep walking and night terrors during intervention (Durand & Christodulu, 2004). This may have been because the sleep restriction programme disrupted the participant's NREM phases, resulting in increased parasomnias (Durand & Christodulu, 2004).

Christodulu and Durand (2004) also tested the effectiveness of sleep restriction in addition to sleep hygiene. Participants on the autism spectrum included a 2.5-year-old girl with PDD and a boy with autism aged 4 years. Sleep disturbances included NWs, and disruptive behaviour at bedtime and during wakings. Following intervention, bedtime disturbances and NW reduced and parent satisfaction with their child's sleep increased. However, sleep quantity was lower following intervention than before implementation of the sleep restriction programme; in fact, the amount of sleep received by these participants would not be considered enough for their age (Christodulu & Durand, 2004). Nevertheless, sleep quality improved. As with bedtime fading, further research is needed to establish the efficacy of sleep restriction for children and adolescents with ASD.

Scheduled awakening. Scheduled awakening procedures are only applicable to children with NW as opposed to sleep onset issues (Owens, France, & Wiggs, 1999). Scheduled

awakening involves parents purposefully waking their child approximately 15 minutes before their usual spontaneous awakenings would be due to occur (Turner & Johnson, 2013). The time periods between scheduled awakenings are increased until the child is able to sleep through the night without waking (Owens et al., 1999). McGarr and Hovel (1980) suggest independent reinitiation of sleep is shaped and reinforced via successive approximations to the desired behaviour, in that parent attention is supplied contingent on being woken from sleep without distress expression (e.g., crying), instead of for spontaneous waking and crying, and the child is woken after increasingly longer sleep periods. The scheduled awakening procedure may also enable young people to practice returning to sleep with discriminative stimuli for the bed environment present, but without the disruption and overshadowing of distress (N. M. Blampied, personal communication, February 27, 2020). In this case, it is critical parents are as non-intrusive as possible to ensure parent-supplied stimuli do not become controlling stimuli for falling asleep (Blampied, 2013a). The efficacy of scheduled awakenings for young people with ASD has not been established; as yet it has only been applied in the treatment of night terrors (a parasomnia; Durand, 2002).

Chronotherapy. Chronotherapy is another behavioural treatment that can be utilised in the treatment of sleep disturbances for children who have a disturbed circadian sleep/wake cycle (Owens et al., 1999). During chronotherapy children are put to bed at a time which is likely to result in rapid sleep onset. Each night the child's bedtime is delayed by 2 hours until the child can fall asleep quickly at an appropriate time (Kodak & Piazza, 2008). Throughout intervention the child's regular wake schedule is maintained (Vriend et al., 2011). This technique has been applied to one child with ASD in an inpatient unit (Vriend et al., 2011). Piazza, Hagopian, Hughes, and Fisher (1998) successfully increased sleep quality while reducing NWs and SOL in an 8-year-old girl with non-verbal ASD. As the participant typically fell asleep at 3:30am, she was placed in bed at this time when treatment began. Her bedtime was delayed by 2 hours each night for the next 8 nights, then by 1 hour on subsequent nights so her sleep patterns did not extend beyond the target bedtime. The participant's daily schedule was modified to fit with her sleep/wake schedule; consequently, she ate breakfast upon waking regardless of the time of day. Once the participant's target bedtime was reached, she was discharged and her mother implemented a consistent bedtime routine in the home. While effective within an inpatient setting, chronotherapy may not be suitable within a home setting as it involves complete disruption of both the parent and young person's everyday schedule (Kodak & Piazza, 2008).

Further research is needed to establish the efficacy of chronotherapy for children and adolescents with ASD.

Parent education programmes. Behavioural sleep interventions are commonly delivered via parent education programmes. Numerous studies have investigated the effectiveness of parent sleep education programmes/workshops in an effort to ensure treatment delivery is efficient and cost-effective. This intervention approach has typically consisted of two to five group education sessions which introduce parents to a range of behavioural management techniques (e.g., sleep hygiene, circadian sleep/wake scheduling, consistent and sleep-conducive bedtime routines, bedtime fading, graduated extinction) that they can utilise to alleviate sleep problems experienced by their child. Parent education programmes have predominantly been implemented with parents of pre-schoolers, or children under the age of 10. Only one study has included parents of children with ASD up to 12 years of age (Roberts, Smith, & Sherman, 2019).

Large single-case studies evaluating parent education programmes have yielded reports of minimal improvement in parent-reported sleep data (e.g., CSHQ scores remained within the clinical range post-treatment; Reed et al., 2009; Yu et al., 2015). A randomised controlled trial also indicated there was no difference in actigraphy-measured sleep variables pre- and post-treatment between parents who received a sleep education programme and parents who received a general autism education programme (Johnson et al., 2013).

A few studies have evaluated the impact of mode of delivery on treatment outcomes. Adkins et al. (2012) found a sleep education pamphlet alone was not effective in reducing sleep disturbance in young children with ASD. Providing parents with a detailed manual as well as necessary resources (e.g., sleep diaries) has produced mixed results (Malow, MacDonald, Fawkes, Alder, & Katz, 2016). This mode of delivery was related to some improvement in actigraphy-measured and parent reported sleep outcomes, but not across all target behaviours (Malow, Macdonald et al., 2016). Further, three of the nine families in this study found it too difficult to read and implement strategies independent of outside assistance. Moon et al. (2010) supplemented provision of a treatment handbook with weekly phone contact with a therapist and this resulted in small improvements in SOL only. Malow et al. (2014) compared the effectiveness of a parent sleep education programme administered individually or within a group format. Both programmes resulted in a reduction in SOL and total CSHQ scores (although the mean score remained within the clinical range), however there was minimal change in other

sleep variables (e.g., SE, total sleep time). Recently, Roberts and colleagues (2019) compared the effectiveness of a parent sleep education programme administered online versus within a face-to-face group. Parents either received two intervention sessions delivered via online podcasts and internet forums, or in interactive workshops. There was a reduction in CSHQ scores (but they remained in the clinical range) at post-treatment for both groups and minimal change in actigraphy-measured sleep variables. It is noteworthy that there was a high attrition rate for the online programme. Parents in this group indicated they did not like talking to other parents online, whereas parents in the face-to-face group enjoyed the peer support aspect. Overall, results suggest face-to-face parent education sessions may be preferred, although both methods were relatively ineffective.

Parent education programmes may be more effective when the content is tailored to the individual family. Austin, Gordon, and O'Connell (2013) devised individualised multicomponent treatment programmes for families during group workshops. At post-treatment there was a statistically significant reduction in sleep disturbance across participants. Two randomised controlled trials have compared individual parent education sessions with a placebo drug treatment condition (Cortesi, Giannotti, Sebastiani, Panunzi, & Valente, 2012) and care as usual (Papadopoulos et al., 2019). Cortesi et al. (2012) found four sessions relating to cognitive (e.g., parent attributions) and behavioural management of sleep problems was effective in reducing insomnia symptoms for 4- to 10-year-olds with ASD. Interestingly, a combined treatment approach consisting of parent education sessions and melatonin administration was more effective than melatonin or education sessions alone (Cortesi et al., 2012). Papadopoulos et al. (2019) found a significant reduction in CSHQ scores for the intervention group compared with the control group, however the mean total score post-treatment remained within the clinical range.

Currently, there is insufficient evidence to support implementation of parent education alone via any mode of delivery. Treatment outcomes have tended to be based on subjective parent report which is at high risk of bias. Further, sleep improvements have not necessarily been clinically substantive, with many studies reporting mean CSHQ total scores within the clinical range post-treatment. Finally, only one of the preceding studies evaluated parent treatment fidelity, therefore it is unclear whether sleep improvements can be attributed to implementation of the recommendations.

Education programmes for young people. Only one study has implemented a behavioural education programme with young people with ASD. Loring et al. (2016) provided two sleep education sessions to 23 adolescents (11 to 18 years) with ASD and their parents. In the first session a psychologist discussed participants' current sleep patterns and provided recommendations based on assessment data. During the second session, role-plays and modelling were utilised to teach participants relaxation techniques to facilitate sleep onset. Adolescents were reinforced for practicing the relaxation techniques with their parents. When the adolescents were competent to carry out the techniques alone, parental assistance was faded. Information was presented to the adolescents based on their personal preference and formats included verbal, written, or PowerPoint presentation. Visual schedules, reminders, task analyses, scripts, and visual reinforcement systems were employed to support their learning. Post-treatment, parent- and self-report data indicated there were significant reductions in negative cognitions and emotions prior to sleep onset, and improvements in the stability of sleep routines across weekdays and weekends. Actigraphy data illustrated significant improvements in SE and SOL, however, it still took 45 minutes on average for participants to fall asleep. This study demonstrated the feasibility of including adolescents with HFA as intervention agents, as well as the effectiveness of a young person- and parent-implemented behavioural intervention for treating sleep disturbance.

CBT. CBT differs from strict behavioural therapy as it includes both behavioural and cognitively mediated intervention techniques to address environment-behaviour contingencies, as well as underlying cognitive states and processes (Beebe & Risi, 2003; Moree & Davis, 2010). Cognitive behaviour modification programmes typically involve the participants' as the primary change agent (Saloviita & Tuulkari, 2000). CBT necessitates the involvement of the young person experiencing the sleep disturbance through the application of cognitively mediated techniques. Cognitive treatment components are commonly implemented alongside behavioural techniques when treating sleep disturbance in typically developing young people. However, reports of the use of CBT to treat sleep disturbance in young people with ASD is largely absent in the literature.

Johnson et al. (2013) provided examples of optional materials which could be incorporated in their parent education programme to address children's nighttime fears. These included coaching parents to teach their child emotional regulation skills and to implement systematic exposure for specific fears. However, the authors did not confirm whether these additional materials were utilised.

Souders et al. (2017) incorporated anxiety management strategies within an individualised, comprehensive multicomponent treatment plan for an 8-year-old boy with HFA. This child experienced separation anxiety and a fear of the dark, and he subsequently co-slept with a family member. Treatment consisted of providing psychoeducation to all family members; a social story; environmental changes (nightlight, decrease available toys, remove TV from bedroom); addressing sensory needs (increased exercise during day); use of a visual schedule with laminated icons on a Velcro board; relaxing bedtime routine (massage, yoga, and deep breathing); faded bedtime; faded parental presence (child agreed to a parent sitting on a chair in his room until he fell asleep, the chair was gradually moved further away); protective item (pillow wrapped in his mother's pyjamas and sprayed in her perfume); and a bedtime pass (described later) with reinforcement for no pass use to encourage brave behaviour. Both parent-report and actigraphy indicated a significant improvement in sleep variables (including elimination of unwanted co-sleeping), and a reduction in co-morbid anxiety.

In 2019, McCrae and colleagues undertook the first study to evaluate modified CBT for insomnia (CBT-I) with children (6 to 12 years) with ASD. All participants had HFA. Modifications to facilitate application to children with ASD consisted of visual supports, incorporation of special interests, repeated information and more practice opportunities, reduced complexity of strategies, use of concrete language, and metaphors. Eight weekly CBT-I sessions were individually administered to 15 children and their parents. Session content consisted of sleep hygiene, consistent bedtime routine, parent behavioural management strategies (e.g., reinforcement, fading of parent presence), circadian education, introduction to cognitive therapy and relaxation, and management of nighttime fears. Intervention techniques involved both parent-implemented (e.g., bedtime limit setting) and child-implemented (e.g., nightmare re-scripting) components. Self-report, parent-report, and actigraphy revealed treatment resulted in improved SOL, duration of wake-after-sleep-onset (WASO), and total sleep time. These outcomes were maintained at 1-month follow-up. This study highlighted children with HFA could participate in intervention with high procedural integrity, and parent-reported treatment acceptability for CBT-I was high.

Pharmacological treatment. As dysregulation of melatonin levels and circadian rhythmicity may contribute to sleep disturbance in individuals with ASD (Gringras, Nir, Breddy, Frydman-Marom, & Findling, 2017), melatonin is commonly prescribed in clinical settings (Cuomo et al., 2017; Malow, Katz et al., 2016). Alongside behavioural intervention, melatonin is the most evidence-based treatment for ameliorating paediatric sleep disturbance in

ASD (Maras et al., 2018). Specifically, it is linked to significantly reduced SOL and increased total sleep duration (Cuomo et al., 2017; Gringras et al., 2017; Maras et al., 2018; Abdelgadir, Gordon, & Akobeng, 2018). Melatonin may also contribute to improved secondary outcomes for parents, such as increased quality of life (Maras et al., 2018), however the relationship between pharmacological treatment and child wellbeing is inconsistent. Schroder et al. (2019) found children receiving melatonin experienced significant reductions in externalising behaviour (particularly those in the clinical range pre-treatment) but not internalising behaviour. Conversely, Malow, Katz et al. (2016) found pharmacological treatment in general was associated with worse child daytime behaviours and lower quality of life.

Notably, there are no clear recommendations regarding melatonin dosage as existing studies have trialled varying doses (range, 0.1 – 12mg; Abdelgadir et al., 2018). Further, the long-term effectiveness of melatonin is relatively unknown, although, preliminary evidence suggests if consumption is continued it may be effective for up to 1 year in some children (Maras et al., 2018). Overall, melatonin usage is not associated with significant risks or side effects (Abdelgadir et al., 2018; Parker et al., 2019). In addition to melatonin a range of other medications for ASD-related sleep disturbance have been investigated (e.g., porcine secretin, antipsychotic risperidone, and α_2 adrenergic agonist clonidine), however there is limited evidence for these treatments (Cuomo et al., 2017; Malow, Byars et al., 2012). Although melatonin remains the most evidence-based pharmacological treatment (Cuomo et al., 2017; Malow, Byars et al., 2012), currently, there is insufficient evidence to determine the effect of any pharmacological treatment on sleep-interfering behaviour, such as bedtime resistance (Cuomo et al., 2017). Critically, there are also currently no medications which meet approval requirements set by pharmacological regulatory bodies (e.g., the Federal Drug Administration in the USA) for the treatment of paediatric insomnia (Malow, Byars et al., 2012; Maras et al., 2018). Once medical causes for sleep disturbance have been eliminated, current guidelines advocate for the use of behavioural and psychoeducational approaches as the first line of treatment (Beresford et al., 2016).

Alternative therapies. Non-behavioural and non-pharmacological approaches to sleep disturbance may include massage therapy, weighted blankets, white noise, bright light therapy, aromatherapy, exercise, and vitamin supplements (McLay & France, 2016). Very few studies have investigated the efficacy of these interventions for ASD-related sleep disturbance (McLay & France 2016). Massage therapy is the most commonly employed alternative therapy (Malow, Byars et al., 2012), however, currently, there is mixed evidence regarding its effectiveness

(Cuomo et al., 2017; McLay & France, 2016). A few studies have reported positive sleep outcomes following Thai or Qigong massage therapy, although objective measurement of sleep variables was not always evident (McLay & France, 2016). Existing research has found minimal or no significant difference in sleep outcomes following vitamin supplements, aromatherapy, or weighted blankets (Gringras et al., 2014; McLay & France, 2016). There is some evidence for increased physical activity/exercise (aquatic exercise, cycling, ball playing/balance activities) and improved sleep variables in young people with ASD, although differences were not always statistically significant (Brand, Jossen, Holsboer-Trachsler, Pühse, & Gerber, 2015; Oriel, Wood Kanupka, DeLong, & Noel, 2016; Wachob & Lorenzi, 2015). Other alternative therapies have not yet been investigated with young people with ASD, although there is some evidence for their effectiveness in typically developing children and adolescents (France, McLay, Hunter, & France, 2018). Consequently, such techniques warrant further investigation in individuals with ASD.

FBA within Sleep Interventions

As described previously FBA is a method used to ascertain the underlying function of behaviour by analysing recurring patterns of behavioural contingencies. It enables practitioners to identify factors that may be contributing toward a young person's sleep disturbance and intervene accordingly. Although, FBA has consistently been shown to increase treatment effectiveness for a wide variety of behavioural conditions and problems, relatively few studies have utilised FBA to inform sleep interventions for young people with ASD. The following section describes the most recent studies employing FBA within this context.

Didden et al. (2002) conducted a functional assessment in relation to disruptive behaviours exhibited by participants at bedtime. One of the children involved in the study was a 6-year-old boy diagnosed with autism. He co-slept with his mother during the sleep onset period, and engaged in disruptive behaviour, such as calling out and crying, during NWs. In response, the child was allowed to leave his bedroom and spend time with his parents in the living room. Functional assessment results were based on parent interviews and reports of antecedent and consequent events each night, as well as the duration of disruptive behaviour. Results suggested the function of the disruptive behaviour was to gain access to parent attention. Consequently, an extinction procedure was implemented (withdrawal of parent attention), leading to a significant reduction in disruptive behaviours.

Moore (2004) also utilised FBA to ascertain the function of sleep problems demonstrated by a 4-year-old boy with ASD. This participant slept in his parent's room; experienced delayed SOL; woke during the night demanding milk; and rose early in the morning. If co-sleeping and the provision of milk were unmet the child engaged in disruptive behaviour. Parent and teacher interviews, home observations, sleep diaries, and questionnaires were used to uncover the underlying function of the child's sleep-related behaviour. Results demonstrated the function was to gain attention and access to tangible items. A multicomponent intervention including graduated extinction and a social story was effective in eliminating co-sleeping and reducing SOL and NWs.

As described earlier, Weiskop et al. (2001) and Weiskop et al. (2005) conducted unmodified extinction to treat sleep disturbance in children with ASD. Across studies, parent interviews and sleep diary data were used to hypothesise the function of each child's sleep disturbance. FBA hypotheses were then used to provide a rationale for the intervention techniques to the parents. Results of the FBA suggest children's NWs were maintained by inappropriate stimulus control, whereby children required the presence of certain conditions at bedtime (e.g., parent presence) that were absent during NWs. Additionally, each individual's disruptive behaviour was positively reinforced by parents' responses. Subsequently, parent behaviour was negatively reinforced via the elimination of their child's disruptive behaviour. Unmodified extinction resulted in improved sleep related outcomes maintained at 3- and 12-months follow-up.

Another study which utilised FBA procedures was conducted by Friedman and Luiselli (2008). In this study FBA was utilised to understand the excessive daytime sleep experienced by a 13-year-old boy with ASD. Two questionnaires and an antecedent-behaviour-consequence checklist completed after each daytime sleep episode were used to generate a hypothesis about the function of the target behaviour. Results indicated daytime sleep was maintained by automatic reinforcement and escape from non-preferred interactions. The subsequent intervention involved removal of sleep-associated stimuli, such as a comfortable chair, and engaging the child in a stimulating activity or with a preferred object when he appeared sleepy. Additionally, the child received praise when engaging in behaviour other than sleeping. As a result, daytime sleep episodes were eliminated, and this change was maintained at 6-month follow-up.

Jin et al. (2013) conducted an FBA to inform interventions for delayed SOL and frequent NWs experienced by two 9-year-old boys with ASD. A clinical interview (guided by the Sleep Assessment and Treatment Tool [SATT]; Hanley, 2005), parent-report sleep diary data, and video observations were used to form a hypothesis about the function of the sleep-interfering behaviour. Results indicated behaviour was maintained by automatic reinforcement (e.g., consequences resulting from the child's own behaviour) and access to attention and tangibles. Consequently, the interventions involved encouraging engagement in stereotypy and providing access to parent attention and tangible items before bedtime to reduce their reinforcement value post-bedtime, occurrences of stereotypy post-bedtime were interrupted, access to tangible items post-bedtime was eliminated, and parent attention was delivered noncontingently on a time-based schedule and gradually faded. Appropriate sleep associations (available throughout the night) were also established. The preceding treatment components successfully reduced sleep-interfering behaviour, delayed SOL, and NWs. These effects were maintained at follow-up.

Finally, three recent NZ studies highlight the effectiveness of FBA-informed interventions in the treatment of delayed sleep onset, CCs, frequent and extended NWs, unwanted co-sleeping, and sleep-interfering stereotypy (McLay et al., 2017; McLay, France, Blampied et al., 2019; McLay, France, Knight et al., 2019). Across studies a clinical interview (guided by the SATT), parent-reported sleep diaries, and VSG were used to generate hypotheses regarding the function of the sleep-interfering behaviour. Results indicated sleep-interfering behaviour and unwanted co-sleeping were maintained by access to tangibles (e.g., breastmilk), parent social attention, and escape from demands. Consequently, intervention included elimination of access to tangibles and parent attention via modified (e.g., fading of parent presence) and unmodified extinction (e.g., access to putative reinforcer available before bedtime or in the morning only) procedures. Additional treatment components to address inconsistent bedtime routines, increase physiological sleep pressure, and encourage desired behaviour included social stories; implementation of appropriate discriminative stimuli for sleep (e.g., Gro-Clock, https://www.gro-store.com/groclock.html); bedtime fading; sleep restriction; and positive reinforcement for sleep-conducive behaviour. White noise and a squeeze ball were provided as stimulus replacements for parent whispering and hand holding respectively. Within the McLay, France, Blampied et al. (2019) study, participant's sleep-interfering stereotypy (repetitive vocalisations and/or motor movements) was hypothesised to be maintained by automatic reinforcement as well as parent attention. Therefore, interventions consisted of white noise to prevent auditory feedback and unmodified extinction of parent attention to reduce the

reinforcing value of stereotypy. Overall, the interventions were mostly effective in reducing target variables, however results were not maintained for a small number of participants whose parents had stopped engaging in the treatment protocol at 6- and 12-week follow-up (McLay et al., 2017; McLay, France, Knight et al., 2019).

Summary of Behavioural Sleep Interventions

Numerous behavioural approaches to treating sleep problems exist. Many of these techniques are highly effective when applied to typically developing children, especially in the treatment of settling and NW (Vriend et al., 2011; Weiskop et al., 2005). However, less research has assessed the utility and applicability of behavioural sleep interventions for young people with ASD. Further research is necessary to determine the efficacy of the following behavioural sleep treatments, unmodified and modified extinction, bedtime fading, sleep restriction, scheduled awakening, chronotherapy, bright light therapy, parent and youth education, CBT-I, and social stories (McLay & France, 2016; Moss et al., 2014; Vriend et al., 2011).

Notably, most studies in this review relied on parent-report measures (e.g., sleep diaries) to evaluate sleep. In numerous studies, this included parent-report questionnaires (e.g., CSHQ) which rely on retrospective approximations of sleep patterns over time and are at high risk of bias. Only two studies utilised self-report measures (Loring et al., 2016; McCrae et al., 2019). Arguably, self-report measures enable a more accurate understanding of the sleep problem to be obtained. Only young people can comment on their perceived sleep quality (Meltzer & McLaughlin Crabtree, 2015), and parents may not always be aware of their child's actions post-bedtime. Parent-report measures were occasionally supplemented by actigraphy. Actigraphy uses limb movements as a proxy to measure sleep/wake states; its reliance on movement reduces its sensitivity to accurately detect wakefulness if a person remains relatively inactive (Moore, Evans, Hanvey, & Johnson, 2017). Only four studies used VSG (Jin et al., 2013; McLay et al., 2017; McLay, France, Blampied et al., 2019; McLay, France, Knight et al., 2019). Video recordings enable detection of salient information (e.g., topographies of awake behaviour) not necessarily identifiable through other measures. It is paramount studies utilise valid and reliable measures of sleep to effectively evaluate the evidence for behavioural interventions. Triangulation of findings by using a range of measures may be necessary.

Almost all reviewed studies applied behavioural interventions to very young, non-verbal children with ASD. However, sleep disturbance is also a significant problem for older children and adolescents with autism across the spectrum of functioning (Allik et al., 2006; Goldman et

al., 2012; Richdale & Prior, 1995). Adolescents in particular are more likely to experience a delayed sleep phase and are vulnerable to environmental influences which interfere with sleep (Loring et al., 2016). For example, older children have increased access to the internet, more freedom to choose their bedtime, engage in late night activities, and require early rising for school (Loring et al., 2016; Mitru, Millrood, & Mateika, 2002). In a study of over 1800 young people (3 to 18 years) with ASD, those older than 11 years had higher rates of sleep disturbance than any other age group (Goldman et al., 2012). Further, older children and adolescents with ASD and sleep problems have often been experiencing such issues for years. Consequently, further research should assess the efficacy of behavioural sleep interventions for older children and adolescents with ASD.

Furthermore, within the ASD and sleep literature relatively few studies have attempted to address the function of the young person's sleep behaviour. FBA is a well-established technique utilised in the assessment and intervention process of problem behaviour in children with developmental disabilities. Recent studies provide promising evidence regarding the effectiveness of FBA-informed sleep interventions for young people with ASD.

Behavioural sleep interventions can be challenging for families to implement, more so if the child has a developmental disability (Meltzer, 2016). The applicability and effectiveness of such strict and potentially disruptive behaviour interventions when carried out in the family's natural sleep environment needs to be evaluated (Jin et al., 2013). Consequently, further research conducted within home-based settings is necessary (Jin et al., 2013).

A number of studies included in this review did not report follow-up data, or did not evaluate long-term follow-up (i.e., over one-month post-treatment). Given sleep problems persist throughout the lifespan if untreated in people with ASD (Sivertsen et al., 2012), it is critical to understand the long-term effectiveness of behavioural sleep interventions. Future research should investigate the maintenance of such interventions.

Another important aspect to consider when evaluating an intervention is consumer perception of social validity (Callahan et al., 2017). Social validity relates to three key aspects of treatment, all of which are rated by the consumer; these include the social significance/ importance of the treatment goal, treatment acceptability/ appropriateness, and satisfaction with the results (Wolf, 1978). These factors can determine whether consumers will adopt and implement treatment (Callahan et al., 2017). Although young people with ASD have been directly experiencing parent-implemented sleep interventions, no existing study has collected

social validity data from the young people themselves. Kazdin (2000) emphasises all parties implementing and receiving intervention should be involved in the assessment of social validity, including children and young people (Kazdin, 2000). It is therefore critical that future studies evaluate the perception of behavioural sleep interventions held by young people with ASD.

Few studies have investigated the impact of resolving sleep problems through behavioural interventions on the daytime behaviour of participants and existing findings are inconsistent. Five studies found a reduction in behavioural difficulties, (e.g., hyperactivity, response inhibition, irritability, inattention) post-treatment (Loring et al., 2018; Malow et al., 2014; McCrae et al., 2019; Moon et al., 2011; Reed et al., 2009), and one study found no improvement (Moss et al., 2014). DeLeon et al. (2004) found a faded bedtime procedure reduced self-injurious behaviour which had previously occurred within 1 hour of waking for the day. Interestingly, although reduced repetitive behaviour has been found by multiple studies (Malow et al., 2014; McCrae et al., 2019; Reed et al., 2009) in a follow-up study to Loring et al's (2016) study no change in restricted/repetitive behaviours was found (Loring et al., 2018). Malow et al. (2014) and Loring et al. (2018) are the only studies to investigate the impact of an ASD-related behavioural sleep intervention on internalising symptoms. They both found a statistically significant reduction in internalising (anxiety, depression, somatisation) behaviours post-treatment. Given that anxiety may be implicated in the sleep disturbance of young people with ASD, this is a critical variable to investigate. Few behavioural treatment studies have evaluated the impact of child sleep interventions on parental sleep (McCrae et al., 2019), parent stress (Moss et al., 2014), or marital relationship quality (Sanberg et al., 2018), and none have explored the wider impact on parent wellbeing.

For the past six decades, parents have overwhelmingly been the primary intervention agent within the autism and sleep literature. Loring et al. (2016), Souders et al. (2017), and McCrae et al. (2019) are the only published studies which have actively included young people with ASD in the therapeutic process of their own sleep disturbance. These studies demonstrate children and adolescents with HFA have the capacity to participate actively in the assessment, treatment, and evaluation process. Additionally, they indicate a mixture of both parent- and young person-implemented treatment components can resolve sleep disturbance. Further, they suggest cognitively mediated treatment components can be applied to young people with ASD alongside behavioural interventions for sleep. Only six other published studies have engaged children with ASD actively in their behavioural sleep intervention (Austin et al., 2013; Delemere & Dounavi, 2018; Moore, 2004; Weiskop et al., 2001; 2005). In these studies,

involvement was limited to application of a social story, pictorial bedtime routine, or reward system only.

The following literature review describes how sleep interventions for typically developing children and adolescents include young people actively within the treatment process. Application of these techniques to young people on the autism spectrum is then discussed, and a review of relevant interventions including young people with ASD in the therapeutic process is reported.

Including Typically Developing Young People in Cognitive Behavioural Sleep Interventions

Behavioural sleep interventions are commonly employed to treat paediatric insomnia in typically developing children and have a strong evidence base (Meltzer & Mindell, 2014; Owens & Mindell, 2011). Unmodified extinction, graduated extinction, parent education, bedtime fading, positive routines, and scheduled awakenings are considered well-established sleep interventions (Meltzer & Mindell, 2014). Additional techniques (e.g., play-based instruction) are often employed alongside these parent-implemented interventions to support the inclusion of children, even those as young as 2 years of age, in the therapeutic process of sleep (Burke, Kuhn, & Peterson, 2004; Sadeh, Hen-Gal, & Tikotzky, 2008). In addition, a range of young person-implemented cognitive and behavioural treatment components are typically incorporated within sleep interventions for older children and adolescents. CBT-I is considered an efficacious treatment for sleep disturbance in typically developing young people (Åslund, Arnberg, Kanstrup, & Lekander, 2018; Blake, Latham, Blake, & Allen, 2019; Blake, Sheeber, Youssef, Raniti, & Allen, 2017; Sadeh, 2005). Further, young person-implemented treatment components (e.g., psychoeducation, relaxation, and mindfulness) for sleep are highly acceptable to young people (Bei et al., 2013; Blake, Sheeber et al., 2017; Bootzin & Stevens, 2005; de Bruin, Oort, Bögels, & Meijer, 2014). The following section describes a range of young person-implemented components commonly included within sleep interventions for typically developing children and adolescents. The evidence-base is also discussed.

Cognitive strategies.

Psychoeducation. Psychoeducation is a core component of CBT and is commonly employed as the first line of treatment for sleep problems. It involves teaching young people about factors which may be contributing to and maintaining their sleep disturbance (e.g., media

use, caffeine consumption, daytime sleep), as well as improving their understanding of typical sleep patterns and architecture (Meltzer & McLaughlin Crabtree, 2015; Paine & Gradisar, 2011). Psychoeducation has been presented in fun and engaging formats to facilitate application to very young children through to adolescents, such as acting out appropriate behavioural sequences using figures in a doll house (Kahn, Ronen, Apter, & Sadeh, 2017). Within Schlarb, Liddle, and Hautzinger (2011) study they employed adolescents as assistants in a sleep lab, tasked with evaluating the benefits of strategies taught to them by a 'sleep doctor'.

Research suggests school-based sleep education programmes are generally not sufficient to improve sleep in children and adolescents (Blunden, Chapman, & Rigney, 2012). While such programmes increase participants sleep knowledge (e.g., Moseley & Gradisar, 2009) and sleep hygiene awareness (e.g., Blake, Schwartz et al., 2017), they do not address critical family level mechanisms (e.g., parent reinforcement of sleep-interfering behaviour) contributing to youth sleep disturbance. Blunden, Benveniste, and Thompson (2016) suggest family involvement is paramount to instigate sleep behaviour change through education programmes. Nevertheless, young people have reported gaining sleep-related knowledge as part of a comprehensive cognitive behavioural and mindfulness sleep intervention was one of the most helpful aspects (Blake, Sheeber et al., 2017).

Motivational interviewing. Motivational interviewing is a therapeutic technique designed to increase a person's intrinsic motivation and commitment to behaviour change (Grosse Holtforth, & Michalak, 2012). This is achieved through the empathetic exploration of ambivalence (Grosse Holtforth, & Michalak, 2012). Within a sleep context, psychoeducation is provided to young people to highlight the discrepancy between their own sleep and recommended sleep, as well as the importance of sleep health (Cain, Gradisar, & Moseley, 2011; Willgerodt, Kieckhefer, Ward, & Lentz, 2014). Barriers to treatment implementation and relapse prevention are discussed (Cain et al., 2011; Willgerodt et al., 2014). Decisional balance sheets and behavioural experiments can also be used to motivate young people to change their behaviour (Cain et al., 2011).

Cain et al. (2011) applied motivational interviewing techniques in conjunction with sleep education to encourage adolescents to improve their sleep habits. This school-based programme successfully increased participant sleep knowledge and motivation to engage in sleep-conducive behaviour. However, although participants indicated they had tried to change their behaviour (i.e., kept consistent sleep/wake schedules), there was minimal difference in sleep parameters

(e.g., SOL). To enhance the effects of motivational interviewing for sleep disturbance, Cain et al. (2011) suggest treatment should focus on youth confidence and readiness to change sleep behaviour, as opposed to exclusively focusing on the importance of sleep. The group education format and lack of parental involvement may have also impeded treatment effects. However, Willgerodt et al. (2014) demonstrated individualised motivational-based interventions applied to school-aged children and their parents had minimal effect on sleep outcomes. Further research investigating application of motivational interviewing techniques to children and adolescents using methodologically sound approaches is necessary to draw more definitive conclusions regarding its effectiveness.

Cognitive therapy. Cognitive therapy is a fundamental component of CBT-I. Cognitive therapy for insomnia involves identifying and restructuring dysfunctional cognitions maintaining sleep disturbance. Individuals are taught to identify dysfunctional beliefs and attitudes and to challenge and replace these thoughts with helpful cognitions. These techniques have been used successfully with children (e.g., Paine & Gradisar, 2011; Palace & Johnstone, 1989; Stewart & Gordon, 2014) and adolescents (e.g., Bootzin & Stevens, 2005; de Bruin, Oort, Bögels, & Meijer, 2014; Gradisar, Gardner, & Dohnt, 2011; Schlarb et al., 2011). Children can be taught simple and concrete methods (e.g., detective thinking [examination of evidence for and against unhelpful thoughts]) to challenge their cognitions (Paine & Gradisar, 2011; Palace & Johnstone, 1989; Stewart & Gordon, 2014) and facilitate independent coping skills. Therapists within a study by Fehr, Russ, and Ievers-Landis (2016), acted out specific sleep scenarios (e.g., waking at night) during play with 4- to 6-year-old children, and modelled cognitive techniques, such as coping self-talk (e.g., “I’m not alone, I have my teddy bear”; p.308). The therapist and child then brainstormed ideas which could be used to address a dolls pretend sleep difficulties (e.g., think of a good dream) and acted out these techniques together during play (Fehr et al., 2016).

While, recent literature reviews and meta-analyses provide support for the use of CBT-I with children and adolescents (Åslund et al., 2018; Blake, Sheeber et al., 2017; Ma, Shi, & Deng, 2018), few studies have investigated which treatment components are integral to its success. Adult studies show reduced dysfunctional and maladaptive beliefs and attitudes regarding sleep are associated with improved sleep outcomes following CBT-I (Schwartz & Carney, 2012). This finding has also been demonstrated with adolescents; Blake, Schwartz et al. (2017) showed improvements in cognitive arousal were related to improvements in sleep quality, suggesting cognitive therapy may be a critical component of CBT-I.

Mindfulness. Mindfulness is a Third Wave cognitive behavioural therapy which involves focusing one's attention on specific stimuli in the present moment, without judgement (Segal, Williams, & Teasdale, 2002). Like cognitive therapy, effective application of mindfulness is thought to reduce sleep disturbance by addressing cognitive processes underlying hyperarousal (e.g., racing thoughts) and facilitating relaxation (Bootzin & Stevens, 2005). Mindfulness is increasingly being incorporated in cognitive behavioural interventions for insomnia. Research shows increased frequency of mindfulness-based practice is related to improved sleep outcomes (Britton et al., 2010; Gradisar et al., 2011). However, only one study has investigated the independent effect of mindfulness on sleep disturbance in adolescence (Bartel, Huang, Maddock, Williamson, & Gradisar, 2018). Bartel et al. (2018) found use of a 15 min guided mindfulness body scan (sequentially drawing awareness to sections of the body) three times a week was effective in reducing prolonged sleep latency experienced by adolescents. However, this intervention technique did not reduce cognitive-emotional arousal, suggesting application of additional components alongside mindfulness may be necessary (Bartel et al., 2018). Nonetheless, adolescent participants within studies by Bei et al. (2013) and Blake, Sheeber et al. (2017) rated mindfulness treatment components as very helpful. As yet, there has not been enough research to determine the evidence base of mindfulness for insomnia in children.

Imagery. Imagery control is the ability to manage mental images (Schmidt, Harvey, & Van der Linden, 2011). Given individuals experiencing insomnia may experience sleep-interfering verbal thoughts or mental images (e.g., ruminating on the day's events), imagery may helpfully redirect such cognitions. It is commonly applied within CBT-I alongside other relaxation strategies, such as muscle relaxation and deep breathing, to address nighttime fears (Blake, Sheeber et al., 2017). For example, McMenamy & Katz (1989) taught 4- to 5-year-old children to imagine pleasant scenes, as well as use relaxation skills, and brave statements (e.g., "I can take care of myself in bed at night"; p. 146) at bedtime. This intervention resulted in reduced nighttime fear, however bedtime resistance still occurred on some nights. Another study which applied imagery in conjunction with muscle relaxation and hypnosis was effective in reducing sleep disturbance in a large group of children and adolescents (Anbar & Slothower, 2006).

Although the effectiveness of imagery alone to treat insomnia has not been investigated with children or adolescents, research with adults indicate promising results. For example, in Harvey and Payne's (2002) study, adults with insomnia who were instructed to imagine a

pleasant and relaxing scene fell asleep quicker and experienced fewer unwanted thoughts at bedtime than adults with insomnia who were given general instructions to distract themselves, or no instructions. Further, Blake, Schwartz et al. (2017) suggest strategies which can reduce purported hyperarousal may be active treatment components within CBT-I for youth.

An alternative technique termed, imagery rehearsal, can be used to replace frightening dream content with nonthreatening content through visualisation (Meltzer & McLaughlin Crabtree, 2015). Children and young people are instructed to re-script their nightmare with alternative content while awake, to reduce associated fear and anxiety, and facilitate mastery and control of dream content. For example, in Palace and Johnston's (1989) study, a 10-year-old male re-scripted nightmare content by imagining he was a superhero able to rescue individuals within the dream from perceived danger. Imagery rehearsal and relaxation instruction were successful in eliminating this participant's nightmares and increasing independent sleeping practices (Palace & Johnston, 1989). Krakow et al. (2001) also successfully reduced nightmare frequency in nine adolescent females following implementation of an imagery rehearsal workshop. Overall, very few studies have applied imagery rehearsal to children and adolescents in the treatment of nightmares, therefore, there is currently insufficient evidence to support the use of imagery rehearsal with this population (Kuhn & Elliot, 2003).

Protective items. Protective items can help children manage nighttime fears (Kushnir & Sadeh, 2012; Sadeh et al., 2008). These items provide an appropriate discriminative stimulus for sleep and can be used to replace parent-supplied discriminative stimuli (e.g., parent presence) which may not be available throughout the night, or those non-conducive to sleep (e.g., iPad). In a large study conducted by Kushnir and Sadeh (2012), 104 children (4 to 6 years) were provided with a soft toy dog (Huggy Puppy) and either told a backstory which encouraged them to care for the dog, or were informed Huggy Puppy was a nighttime companion to protect them and help them overcome their fears. Both methods were effective in reducing nighttime fears and improving sleep quality. Delivering care to the Huggy Puppy was thought to enhance the child's self-esteem and distract them from their own distress, and the companion Huggy Puppy was thought to facilitate self-regulation. Other 'protective' items could include a Dream Catcher, 'Monster Spray', or a superhero stuffed toy (Mindell & Owens, 2015). Protective items are often incorporated within multicomponent interventions for sleep disturbance and there is promising evidence supporting this technique in the treatment of nighttime fears experienced by young children.

Bibliotherapy. Bibliotherapy presents evidence-based treatment strategies in a storybook format and has been utilised to address nighttime fears and unwanted co-sleeping in typically developing children (Lewis, Amatya, Coffman, & Ollendick, 2015; Rafihi-Ferreira, Silvares, Asbahr, & Ollendick, 2018). Rafihi-Ferreira et al. (2018) found implementation of bibliotherapy in conjunction with a protective item was effective in reducing unwanted co-sleeping and nighttime fears in 4- to 6-year-old children. In this study parents were instructed to read *Sleeping with Rafi: Good night my child*, nightly to their child. The children were given a stuffed kangaroo doll and encouraged to sleep independently. The story outlined an appropriate sleep routine and modelled independent sleeping despite the young kangaroo's fear of sleeping alone. The story emphasised the safety of the kangaroo although it was physically distant from its parents. The authors suggested the participants likely identified with Rafi the kangaroo and subsequently attempted to imitate its behaviour. Lewis et al. (2015) used a similar technique with 5- to 7-year-olds which also reduced nighttime fears and unwanted co-sleeping. In this study, parents were instructed to read one chapter of *Uncle Lightfoot: Flip That Switch* to their child every evening and encourage them to engage in the exposure exercises/games which were detailed in the story (e.g., finding toys in the dark). Initial findings suggest bibliotherapy may be an effective tool to encourage young children to engage in self-directed cognitive behavioural strategies. Perhaps unsurprisingly, parents thought the use of child inclusive techniques, such as stories and dolls/ soft toys, contributed to the young person's motivation and interest to engage in therapy (Rafihi-Ferreira et al., 2018). Of note, social stories differ from bibliotherapy in that they are primarily tailored to the individual's abilities (e.g., appropriate vocabulary, font size, length), written according to a specific format (e.g., include descriptive, directive, and perspective statements), and exclusively use first- and/or third-person statements as well as literal and accurate language (e.g., the statement "I can use my phone before bedtime" is used instead of "I will use my phone before bedtime" as the latter may not always be true; Gray & Garand, 1993; Gray, 2010; Gray, 2013).

Behavioural components.

Bedtime routine. Implementation of a consistent and non-stimulating bedtime routine can promote relaxation, strengthen cues for sleep, and help entrain an individual's circadian rhythm to cues in the external environment (Jan et al., 2008). Positive bedtime routines are a well-established treatment method for sleep disturbance in typically developing children and adolescents and a core aspect of sleep hygiene psychoeducation (Meltzer & Mindell, 2014; Mindell, Kuhn, Lewin, Meltzer, & Sadeh, 2006). A number of techniques can be utilised to

facilitate application to young people. For example, bedtime charts or picture schedules can present the young person's bedtime routine pictorially. Tangible objects, such as a toothbrush, can be permanently affixed to the chart. These strategies enable young people to understand each step in their bedtime routine (Meltzer & McLaughlin Crabtree, 2015). Further, caregivers can prevent negotiation tactics by referring back to the chart if their child asks for something which is not part of their routine (e.g., extra time on their device; Meltzer & McLaughlin Crabtree, 2015).

Stimulus control. Stimulus control consists of establishing and strengthening discriminative stimuli that reliably provide cues for sleep-conducive behaviour and sleep onset (e.g., dark room) and eliminating cues which are incompatible with sleep (e.g., sleep-interfering activities, hyperarousal; Bootzin & Stevens, 2005). Stimulus control is considered a well-established sleep intervention for typically developing young people (Meltzer & Mindell, 2014) and is well-accepted (Blake, Sheeber et al., 2017). This treatment strategy has been employed regularly in the treatment of insomnia in children (Paine & Gradisar, 2011) and adolescents (Bootzin & Stevens, 2005; Clarke et al., 2015; de Bruin et al., 2014; de Bruin, Bögels, Oort, & Meijer, 2015; Hendricks, Ward, Grodin, & Slifer., 2014; Schlarb et al., 2011; Roeser, Schwerdtle, Kübler, & Schlarb, 2016). Stimulus control training involves teaching young people to alter their sleep/wake schedules (i.e., delay their bedtime, rise at a consistent time) to facilitate sleep onset and increase homeostatic sleep pressure, restrict the use of stimulating activities in bed (e.g., electronic device use), and to leave their bed to engage in a non-stimulating activity until they feel tired if unable to initiate sleep in 30 minutes (Hendricks et al., 2014). Such techniques strengthen bed-specific discriminative stimuli controlling sleep onset, thus establishing the bed as a strong discriminative stimulus for sleep. There is significant overlap between positive bedtime routines, sleep hygiene, stimulus control, and bedtime fading with response-cost, as utilised with younger children.

For young people unable to discriminate between sleep/wake times, special night lights (e.g., a Gro-clock) can be used to establish a stimulus control system. These night lights provide a visual cue (e.g., star/sun on the clock face) to young people letting them know when it is bedtime and when it is acceptable to rise for the day (Meltzer & McLaughlin Crabtree, 2015). Young people are instructed to remain in bed until the symbol for daytime appears and are typically rewarded for doing so. While these tools are favoured anecdotally by parents, their use has not been documented extensively in the literature. No studies were identified which utilised a visual indicator of sleep/wake times with typically developing young people and only one such

study was identified within the autism and sleep literature. In this study, McLay, France, Knight et al. (2019) incorporated a Gro-clock within a multicomponent behavioural treatment for 2- to 5-year-olds with ASD experiencing unwanted co-sleeping in addition to difficulty settling and maintaining sleep.

Relaxation. Relaxation training is often incorporated in multicomponent CBT-I treatment for typically developing children, adolescents, and adults. Relaxation strategies are thought to facilitate sleep initiation and maintenance by alleviating cognitive and/or physiological arousal (Didden et al., 2014; Reaven, 2009). However, given relaxation instruction is rarely employed independently of other cognitive behavioural strategies, its individual contribution to sleep treatment is unknown. Nevertheless, initial evidence suggests adolescent participants perceive learning relaxation skills to be a helpful addition to their sleep treatment (Bootzin & Stevens, 2005; de Bruin et al., 2014). Further, providing relaxation instruction to young people can give them the confidence and skills to manage nighttime fears independently (Pincus, Weiner, & Friedman, 2012).

Common relaxation techniques utilised within paediatric sleep interventions include progressive muscle relaxation (PMR) and deep breathing (Bootzin & Stevens, 2005; de Bruin et al., 2014; 2015; Roeser et al., 2016; Schlarb et al., 2011). PMR includes purposefully tensing and releasing different muscle groups throughout the body, whereas deep breathing involves taking slow, controlled breaths through the diaphragm. Singly applied, compared to other relaxation strategies, PMR has the most evidence for treating adult insomnia (Taylor & Roane, 2010). However, combining somatic management techniques with guided imagery enables both cognitive and behavioural factors implicated in paediatric sleep disturbance to be addressed. Relaxation strategies can be taught to young children through fun and engaging methods, such as incorporating a soft toy (Schlarb, Velten-Schurian, Poets, & Hautzinger, 2010). The addition of a relaxation script can encourage regular practice and provides the young person a resource to facilitate independent application (Stewart & Gordon, 2014).

Exposure therapy. Exposure therapy involves gradually exposing a young person to feared stimuli (e.g., darkness, sleeping alone) to promote feelings of safety in the repeated absence of aversive consequences. Exposure therapy is a well-established evidence-based component of CBT for paediatric anxiety (Böhnlein et al., 2020; Whiteside et al., 2020). The effectiveness of exposure therapy within the treatment of nighttime fears has also been consistently demonstrated (Kahn et al., 2017; Paine & Gradisar, 2011; Pincus et al., 2012;

Stewart & Gordon, 2014). Exposure therapy is typically implemented once young people have developed relaxation skills to manage feared situations (Meltzer & Mindell, 2015). With younger children, exposure exercises can be implemented through play, such as seeking an object in progressively darker environments (Huebner, 2008; Kahn et al., 2017). Additionally, parents can be appointed as 'fear-fighting coaches' to help their child complete exposure exercises (Stewart & Gordon, 2014)

Bedtime pass. A bedtime pass can be used to address bedtime resistance (e.g., CCs), as well as facilitate independent sleep throughout the night. Bedtime passes are small cards which children can use to make a specific number of negotiated requests for parental attention once in bed (Meltzer & McLaughlin Crabtree, 2015; Mindell & Owens, 2015). Traditionally, once children have used their allocated passes, further bids for parental attention are ignored (modified extinction; Meltzer & McLaughlin Crabtree, 2015). To encourage children to stay in their bedroom post-bedtime rewards can be earned for settling without use of the pass. Multiple studies have demonstrated its effectiveness at reducing bedtime disturbance and sleep onset for typically developing children as well as demonstrating high parental ratings of treatment acceptability (Freeman, 2006; Friman et al., 1999; Moore, Friman, Fruzzetti, & MacAleese, 2007).

Reinforcement. Reward systems describe an operant conditioning process whereby children are provided with positive reinforcement contingent on appropriate sleep behaviour (Mindell & Owens, 2015). Reinforcement procedures are commonly included within CBT-I interventions to encourage children to engage in sleep-conducive behaviour. Reinforcement can be social (e.g., in the form of praise), or tangible (e.g., small item) and is delivered as soon as possible after the desired behaviour occurs (Mindell & Owens, 2015). Creative application of reward systems can encourage child buy-in. For example, in Burke and colleagues' (2004) study parents read a storybook to their child nightly which explained that a 'sleep fairy' would place a small prize (e.g., bookmark, hair ribbon) underneath their pillow if they slept through the night without disturbance. Burke et al. (2004) found implementation of bibliotherapy (which took advantage of children's magical thinking abilities) and reinforcement resulted in reduced bedtime resistance and NWs in children aged 4 to 7 years. Once children reached mastery criteria, reinforcement was provided intermittently.

Application of Procedures to Young People with ASD

Treatment challenges specific to working with young people with ASD.

Although cognitive behavioural treatment strategies are effective when applied with typically developing young people, debate exists about their applicability to young people with ASD (Moree & Davis, 2010). Core features of autism symptomology, such as marked difficulties with social communication, can challenge the provision of many different forms of psychological therapy. Effective implementation of cognitive and behavioural treatment techniques requires certain abilities which can be impaired in individuals with ASD (Lickel, MacLean, Blakeley-Smith & Hepburn, 2012).

Important skills to facilitate CBT-I include perspective taking, self-reflection, metacognition, causal reasoning, sufficient memory, emotion recognition, interoception, and expressive and receptive language capabilities (Lickel et al., 2012). Perspective taking may enable young people to understand the impact of their sleep disturbance on other family members, as well as recognise that their sleep behaviour is atypical. Self-reflection and metacognition skills would enable individuals to identify their sleep-interfering thoughts, facilitating thought disputation. Further, causal reasoning is likely to facilitate recognition of the importance of their own thoughts, feelings, and behaviour on sleep-related outcomes. Additionally, it is critical for young people to be able to identify internal body states (interoception; e.g., tiredness, rapid heartbeat) in order to appropriately self-regulate. Individuals also need to be able to remember their sleep patterns and behaviour and have the ability to communicate these. Receptive and expressive language capabilities are necessary to facilitate discussion and comprehension of sleep related topics involved in the therapeutic process. However, all of these processes can be challenging for people with ASD (Attwood & Scarpa, 2013). The following section discusses ASD-related characteristics and the challenges these may pose to therapeutic processes.

Theory of mind (TOM). TOM refers to the ability to imagine or understand the mental states of others to make sense of their behaviour (Attwood, 2007; Baron-Cohen et al., 2005). It is theorised that this is particularly challenging for individuals with ASD and may underlie social communication difficulties (Attwood, 2007; Baron-Cohen et al., 2005). TOM impairments can prevent individuals from understanding the perspectives of others, or even that others may hold a different perspective to their own (Granpeesheh & Tarbox, 2008). It can also inhibit one's ability to predict the intentions of others (e.g.., detection of irony and sarcasm) and subsequently respond appropriately (Scheeren, de Rosnay, Koot & Begeer, 2013; Zalla, Miele,

Leboyer & Metcalfe, 2015). Such challenges may relate to difficulties establishing and maintaining reciprocity during social interactions (Tager-Flusberg, Paul, & Lord, 2005). For example, when talking on the phone a young person with ASD may begin discussing events the listener cannot perceive and remain unaware the listener does not have the same knowledge or comprehension as they do. During conversations individuals with ASD may struggle to contribute evenly, take turns, and to offer appropriate or relevant information (Parsons, Cordier, Munro, Joosten, & Speyer, 2017). Instead, they may exclusively engage in lengthy monologues regarding topics of special interest to them. Non-adherence to social conventions of conversations can limit rapport building and therapeutic discussions.

TOM impairments not only affect a person's ability to think about the thoughts and feelings of others but may contribute to difficulty engaging in metacognitive processes (i.e., thinking about one's own thoughts and feelings; Attwood, 2003; Grainger, Williams, & Lind, 2014). Consequently, it can be even more challenging for people with ASD to identify and regulate unhelpful cognitions and unpleasant emotions. Young people with ASD are more likely to experience alexithymia (difficulty identifying, differentiating, and articulating emotions) than typically developing youth (Roberts-Collins, Mahoney-Davies, Russell, Booth, & Loades, 2018). TOM and executive function impairments mean individuals on the autism spectrum are particularly vulnerable to experiencing maladaptive cognitions and dysfunctional beliefs, as well as emotional dysregulation (Attwood, 2007).

Cognitive and executive functioning. There is wide variability in cognitive and adaptive functioning across the spectrum, with some individuals experiencing comorbid ID and others extremely high IQs. Further, individuals with ASD tend to have diverse cognitive profiles, demonstrating strengths and weaknesses in a range of different areas, such as spatial processing versus verbal comprehension (Atwood & Scarpa, 2013). Challenges related to executive functioning are also common. Individuals with ASD can demonstrate cognitive inflexibility, difficulty regulating attention, and compromised planning and organisational capabilities (Attwood & Scarpa, 2013; Beebe & Risi, 2003; Geurts, Sinzig, Booth, & Happé, 2014; Ho, Stephenson & Carter, 2015; Mercado, Kratz, Frank, Wolensky, & Kerns, 2018). Consequently, it may be harder to reconceptualise problems and generate alternative thoughts and behaviour, as well as sustain attention on therapeutic stimuli. Further, cognitive inflexibility may relate to preference for routine and rituals (Beebe & Risi, 2003), contributing to resistance to change during the course of therapy. Lastly, difficulty conceptualising another person's point of view

(TOM) in conjunction with cognitive inflexibility, can impede negotiation or compromise and contribute to conflictual interactions.

Sensory processing. Atypical sensory processing is a core feature of ASD. Individuals with ASD may experience hypo (e.g., nonresponsive) and hyper (e.g., over-stimulated) responsivity to stimuli. Poor integration of sensory input, or sensory modulation difficulties can inhibit self-regulation. Interoception refers to the conscious awareness of internal body experiences. Awareness of internal states is necessary to effectively self-regulate. For example, a person needs to be able to notice bodily sensations (e.g., droopy eyelids, reduced energy) and interpret these correctly (e.g., tiredness) in order to resolve such feelings (e.g., sleep). Preliminary research suggests interoceptive awareness may be impaired in some individuals with ASD (DuBois, Ameis, Lai, Casanova, & Desarkar, 2016). They may feel overwhelmed by internal signals, may not notice internal body signals until they are intense, or they may not be able to discriminate between sensations (Mahler, 2017). Consequently, individuals with ASD may struggle to monitor and effectively manage internal states related to physiological processes (e.g., fatigue) or emotions (e.g., anxiety).

Language and communication. Individuals on the autism spectrum have a diverse range of speech and language profiles, which may affect comprehension and expression during therapy (Attwood, 2007; Ministries of Health & Education, 2016). Youth with ASD can experience significant delays in language development, or may even be non-verbal, through to demonstrating high verbal IQs and engaging in exceptionally precise and formal speech (Tager-Flusberg et al., 2005). Variations in speech and language abilities are often related to intellectual ability. For example, young people with ASD and little to no speech tend to have a significant ID (Sigafoos, Schlosser, O'Reilly, & Lancioni, 2009).

ASD is associated with a number of speech and language features. For example, echolalia, which involves echoing another person's words/phrases, is commonly engaged in by people with ASD. Unusual paralinguistic features (e.g., monotony) and behaviour (e.g., lack of eye contact), are also characteristic of people with ASD (Tager-Flusberg et al., 2005; Landa, 2007). For example, they may not use grammatical or affective prosody, such as varying their pitch, tone, and speech rate to convey their feelings, draw the listeners attention to important information, or indicate they are asking a question (Tager-Flusberg et al., 2005). Consequently, comprehensibility is compromised.

Because social communication impairment is a core feature of ASD, difficulties in this area are evident even when linguistic skills (e.g., vocabulary) are intact. This includes difficulty responding contingently to another's remarks during conversation, not accounting for the situational context of spoken language, literal interpretations of speech, and difficulty understanding and engaging in appropriate non-verbal communication (e.g. gestures, facial expressions; Tager-Flusberg et al., 2005). Further, some individuals with ASD may largely engage in conversation for functional (e.g., request) instead of social purposes (Sigafoos et al., 2009).

***Social motivation and anxiety*.** In general, humans are predisposed to orient towards a social world, seek out and find pleasure in social interactions, and work to maintain social relationships (Chevallier, Kohls, Troiani, Brodkin & Schultz, 2012). However, individuals with ASD may be less likely to orient towards social stimuli (Chevallier et al., 2012). Social motivation, approach, and reward varies amongst individuals with ASD (White et al., 2018). Some young people with ASD find social rewards (e.g., praise), less reinforcing than typically developing children, and are less likely to initiate interactions (Chevallier et al., 2012). On the other hand, individuals with HFA may be highly motivated to engage in social interaction, however the nuances of social engagement are not necessarily intuitive to them (Carter, Ornstein Davis, Klin, & Volkmar, 2005).

Inhibited social interactions with individuals with ASD may reflect social anxiety as opposed to low social motivation or lack of interest (White & Roberson-Nay, 2009). It is common for individuals with ASD to experience social anxiety, leading to avoidance of or distress in social situations (Mercado et al., 2018; White, Bray, & Ollendick, 2012). Individuals with HFA who have insight into their social communication difficulties may experience fear of humiliation and rejection given the possibility of misinterpreting social cues (White & Roberson-Nay, 2009). In a qualitative study including adults with diagnoses of Asperger's and HFA, participants described dreading casual conversations with others which did not follow a predictable set of rules and required improvised responses (Müller, Schuler & Yates, 2008). Social anxiety may further exacerbate social communication challenges (White & Roberson-Nay, 2009). As many psychological or talking therapies necessitate social interaction, they can be challenging for young people with social communication difficulties (Cooper, Loades, & Russell, 2018). Further, social communication difficulties can interfere with the therapeutic relationship (Cooper et al., 2018); a core component of effective therapy (Albaum, Tablon, Roudbarani, & Weiss, 2019).

Cognitive and behavioural therapy techniques need to accommodate the unique profile of abilities and features consistent with a diagnosis of ASD (Attwood & Scarpa, 2013). As with all young people, it is necessary to account for their individual development, cognitive abilities, interests, strengths, and difficulties (Moree & Davis, 2010). The heterogeneity of ASD means individuals with this diagnosis may experience challenges in some of the aforementioned areas but not others, and to varying degrees. The presence of any of these difficulties may interfere with the therapeutic process. In order to involve young people with ASD in the therapeutic process adaptations to assessment and intervention strategies applied with typically developing young people are necessary.

Modifications to Include Young People with ASD in the Therapeutic Process

The preceding section illustrated core features of ASD can challenge treatment delivery, however, young people with developmental disabilities are able to benefit from standard therapeutic interventions if appropriate modifications are carried out (Reaven, 2009; Cooper et al., 2018). The following section highlights the numerous techniques which can be utilised to facilitate the inclusion and active participation of young people with ASD during assessment, intervention, and evaluation procedures. Such modifications can mitigate potential challenges.

Communication. Although meeting the communication needs of participants with disabilities can be challenging, the importance of developing effective communication has been a key theme within existing studies that include children with disabilities as active agents (Bailey et al., 2015). During interviews with three young people with PDD and Asperger's disorder, interviewers found rephrasing questions to suit the young person's understanding, clarifying the accuracy of the young person's interpretation, and providing an extended period of time after asking a question facilitated information gathering. Use of closed questions, forced choice, or multiple-choice options have also been shown to facilitate communication with young people with ASD (Harrington et al., 2013; Walters, Loades, & Russell, 2016). Open questions (e.g., tell me about your sleep last night) may overwhelm individuals with ASD, particularly if TOM challenges impact their ability to understand the intention behind the question (Attwood, 2007).

Communicating a range of response options to young people with ASD can help facilitate flexible thinking and may prompt them to generate alterative responses also (Attwood & Scarpa, 2013). This is a common modification to CBT for young people with ASD (Walters et al., 2016). For example, a list of unhelpful/helpful cognitions is provided to participants to

choose from, instead of having to generate their own (Walters et al., 2016). Further, use of pictorial response options can minimise word-retrieval difficulties experienced by some people with ASD (Attwood, 2003).

Visual resources are commonly used to facilitate communication with young people with ASD. It can be challenging for individuals with ASD to communicate their cognitions verbally, especially when those cognitions are pictorial (Attwood, 2003). Young people with ASD may be better able to express their thoughts and emotions through forms of communication other than speaking (e.g., art, music, text; Attwood, 2003). Visual resources, drawing, and tape recordings have been used to engage young people with reduced verbal ability (Bailey et al., 2015). For example, Harrington et al. (2013) used emotion picture cards to enable participants with limited verbal ability to express their feelings. Providing information in a range of formats (e.g., verbally, pictorially, and written) is thought to enhance comprehension (Harrington et al., 2013; Kaifas-Tennyson, 2008; Walters et al., 2016) and complementing verbally mediated material with visual aids (e.g., handouts) reduces reliance on memory and social interaction.

Lastly, communication can also be facilitated through recognition of the increased anxiety experienced by some individuals on the autism spectrum and adapting techniques accordingly. Beresford et al. (2004) conducted interviews with children with ASD side by side while completing an art activity. This method was thought to alleviate social anxiety by minimising direct face-to-face contact (Beresford et al., 2004). Indirect methods of conversation, such as text or email, mean youth with ASD do not have to try and interpret non-verbal communication (e.g. tone, or facial expressions).

Concrete and visual techniques. Concrete and visual techniques are identified extensively in the literature as appropriate strategies to facilitate communication, comprehension, and implementation of treatment strategies with young people with ASD (Burkhart, Knox, & Hunter, 2018; White et al., 2018). Communication methods with a temporal structure (e.g., gestures) are more difficult for individuals with ASD to comprehend due to their transient nature and the necessary ability to process information sequentially (Noens & van Berckelaer-Onnes, 2004). Communication methods which are concrete and contain a spatial structure, such as objects, photographs, and written text, are preferable (Noens & van Berckelaer-Onnes, 2004). Accordingly, social stories are often used to introduce and explain CBT concepts (e.g., problem solving) to young people with ASD (Walters et al., 2016). Visual

cues and schedules (e.g., a pictorial bedtime) are used to compensate for deficits in executive functioning (e.g., working memory, planning) and enable individuals with neurodevelopmental disorders to complete tasks independently, facilitating self-management of their own behaviour (Saloviita & Tuulkari, 2000). They can also facilitate transitions between activities (e.g., during therapy, steps in a routine; Dunlap, Iovannone, & Kincaid, 2008).

Research suggests less emphasis on abstract language or ideas are critical modifications to cognitive behavioural approaches to accommodate the potentially literal mind-set of young people with ASD (Moree & Davis, 2010; Reaven, 2009). In order to teach abstract concepts (e.g., emotions) to young people with ASD a more concrete approach is required (Attwood & Scarpa, 2013; Reaven, 2009; Wood & Schwartzman, 2013). For example, to teach young people how to recognise different emotional experiences, education regarding the associated physical symptoms may be helpful (Rotheram-Fuller & MacMullen, 2011). As young people with ASD can find can it difficult to quantify abstract concepts (e.g., emotion, mood), numerical or pictorial rating scales are commonly used within the CBT and autism literature to enable more accurate assessment (Attwood, 2003; Walters et al., 2016). Perhaps rather counterintuitively, metaphors can also be used to help make some concepts and ideas more concrete and are commonly utilised within CBT for young people with ASD (Attwood & Scarpa, 2013; Walters et al., 2016). For example, practitioners can provide children with a "toolbox" full of techniques or tools they can use to help them in certain scenarios (Attwood & Scarpa, 2013).

Video-based instruction. Observational learning procedures have been used to teach children and adolescents with ASD a range of skills and behaviours (Darden-Brunson, Green, & Goldstein, 2008). In-vivo modelling consists of observing models (e.g., a familiar or unfamiliar peer) complete target behaviour in real time. This practice has been effective in teaching young people with ASD diverse skills and behaviours across a range of contexts (Darden-Brunson et al., 2008). However, there are a number of limitations to in-vivo modelling, which may indicate the use of video modelling (VM) or video self-modelling (VSM). VM involves watching a model perform target behaviours through a video format. Alternatively, VSM involves watching the self (target person) perform target behaviour via a video format.

VM and role-play is commonly used to teach cognitive behavioural strategies (e.g., relaxation exercises) to young people with ASD (Keefer, White, Vasa, & Reaven, 2018; Walters et al., 2016). Research shows both VM and VSM are effective techniques to teach new skills and behaviour to children and adolescents with ASD and have high social validity (Bellini &

Akullian, 2007; Cardon, Guimond, & Smith-Treadwell, 2015). There are numerous reasons why VM may be even more effective than in-vivo modelling. Firstly, research suggests VM/VSM lead to greater generalisation of target behaviour across persons, settings, and stimuli, as the target person is already one step removed from the learning environment, compared to in-vivo modelling (Bellini & Akullian, 2007; Charlop-Christy, Le, & Freeman, 2000). Secondly, many young people with ASD enjoy watching videos, therefore, VM/VSM may be more engaging than in-vivo modelling as it is associated with recreation and is novel (Bellini & Akullian, 2007; Charlop-Christy et al., 2000). Consequently, young people with ASD may be more likely to attend to VM, resulting in faster acquisition of targeted behaviour (Charlop-Christy et al., 2000). Thirdly, in-vivo modelling involves more cognitively complex surroundings and may be less engaging for individuals with ASD, whereas VM/VSM use camera angles and zooming to encourage viewers to focus on relevant cues and facilitate learning of the target behaviour (Bellini & Akullian, 2007; Charlop-Christy et al., 2000). Finally, VSM reduces reliance on social communication skills, such as eye contact with a model, that may be difficult and/or anxiety provoking for young people with ASD (Charlop-Christy et al., 2000).

Parent involvement. Young people with ASD may rely more heavily on their parents than their same age counterparts (White, Scarpa & Attwood, 2013). Parent involvement is critical to facilitate the triangulation of information and increase treatment effectiveness and maintenance (Moree & Davis, 2010; Symon & Boettcher, 2008). Attending sessions with parents facilitates the implementation of therapeutic techniques at home and enables external monitoring of compliance (Gradisar et al., 2011; Paine & Gradisar, 2011; Walters et al., 2016). During conversations with a therapist, caregivers are able to clarify whether children's responses are incomplete or inaccurate, and can provide memory prompts (Harrington et al., 2013; Preece & Jordan, 2010). Parental presence may also reduce their child's anxiety during assessment and therapy sessions (Kreslins, Robertson, & Melville, 2015). Additionally, parent involvement may be particularly important when family factors are contributing to the young person's issues (Kreslins et al., 2015). Research shows CBT for anxiety in young people with ASD is more effective when parents are involved, compared with no involvement (Perihan et al., 2019).

Although there are many benefits to parental involvement, parental presence can be problematic when children defer to their caregivers as opposed to attempting communication (Preece & Jordan, 2010). Or if caregivers prohibit participation in therapeutic tasks for fear of provoking anxiety or perceive their child to be incapable of performing the task (Beresford et al., 2004). White and colleagues (2013) argue further research is required which explores the

extent to which parental involvement is needed when working with youth with HFA in particular.

Sensory needs. People with ASD are prone to experiencing sensory modulation difficulties. Consequently, it may be necessary to account for individual sensory processing needs to facilitate engagement in the assessment and therapeutic process. Research suggests numerous modifications can be made to meet such needs, including adjusting the lighting, sounds, smells and seating arrangements within the assessment or therapeutic environment (Attwood & Scarpa, 2013; Cooper et al., 2018). Other adaptations may include providing opportunities for clients to satisfy their desire for sensory stimulation in non-disruptive ways, such as allowing the young person to handle a small item (Attwood & Scarpa, 2013; Cooper et al., 2018).

Structure and routine. People with ASD often have a preference for routine and sameness. Predictable assessment and therapeutic sessions, whereby a particular structure or organisation is adhered to consistently, may alleviate anxiety, facilitate engagement, and support the acquisition of skills and behaviours**.** As cited in Chilvers (2007, p.20) a 17-year-old with Asperger's reported "I can get easily confused by instructions. I like things kept simple and I like to have a routine". Because of executive functioning deficits, therapeutic sessions which include short activities, and are split into smaller components are more likely to be effective for young people with ASD (Attwood & Scarpa, 2013). Young people with ASD also benefit from highly structured sessions with clear expectations (White et al., 2018). Harrington et al. (2013) demonstrated visual schedules which illustrate session structure, can prepare young people with ASD for upcoming content and help maintain attention.

Incorporating special interests. Young people with ASD may have reduced motivation and a limited attention span when focussing on activities outside their specific domains of interest (Wood & Schwartzman, 2013). Young people's interests are often incorporated within cognitive behavioural treatment programmes to enhance their engagement, attention, and comprehension (Attwood & Scarpa, 2013; Walters et al., 2016; White et al., 2018). Assessment tools, treatment resources, and reward systems which are related to special interests can be created (Chilvers, 2007). For example, if a young person has a special interest in a superhero, an illustrated example of how this character manages certain situations could be provided (Attwood & Scarpa, 2013). However, while using such approaches appropriately can enhance motivation and willingness to engage in treatment, it is important practitioners strike a balance between

incorporating child interests for therapeutic gains, and encouraging potentially problematic fixations (Moree & Davis, 2010). Mercado et al. (2018) suggest using a visual chart or agenda to specify when special interests can be discussed (Mercado et al., 2018).

Overall, research suggests many modifications to traditional intervention approaches can be utilised to enhance their application to young people with ASD. Cognitive behavioural techniques in particular can be easily individualised to suit a person's neurodevelopmental needs and presentation. Further, any ratio of cognitive and behavioural components can be included as required.

Therapeutic content needs to be specifically tailored to meet the needs of individuals with ASD (Kreslins et al., 2015). ASD is a heterogeneous condition and each individual presenting for support is unique. Therefore, modifications to support the inclusion of youth with ASD in the therapeutic process need to be guided by the individual, as opposed to homogenous assumptions about people with ASD in general. Within a study conducted by Beresford et al. (2004), two children with ASD were able to answer abstract questions, and parents were surprised at the extended length of time their children were able to spend with the researcher. Consequently, it is important to approach young person involvement flexibly to allow for diverse abilities (Bailey et al., 2015; Reaven, 2009).

Interventions Including Young People with ASD in the Therapeutic Process

Due to the paucity of research involving children and adolescents with ASD in the therapeutic process of sleep, a wider review of inclusive interventions for diverse issues experienced by young people with ASD was conducted. Specifically, a review of self-management, mindfulness, and CBT procedures was carried out. These therapies were selected based on their similarity to sleep interventions applied to typically developing young people, as well as their capacity to actively include young people with ASD in the therapeutic process. Review articles exploring the effectiveness of interventions targeting challenging behaviour and anxiety in young people with ASD were included. These two issues were considered the most relevant as they are common co-existing challenges faced by young people with ASD. Further, participants within the current study are likely to exhibit a similar comorbid profile given the contribution of challenging behaviour and anxiety to sleep problems.

Databases searched included: PsycInfo; Psychology and Behavioral Sciences Collection; ERIC; and Embase. The searches combined the terms: "autism spectrum disorder",

"ASD", "Asperger syndrome", "pervasive developmental disorder", "neurodevelopmental disorder", "cognitive behavio*r therapy", "CBT", "mindfulness", "mindfulness based intervention", "MBI", "mindfulness based stress reduction", "MBSR", "self-management", "self-monitoring", "self-consequation", "anxiety", "challenging behavio*r", "disruptive behavio*r", internali*ing behavio*r", "externali*ing behavio*r" "evaluation", "effectiveness", and "efficacy". The review, presented below, focuses only on the most recent and relevant research and presents results related to the primary outcomes. Studies were included that have been conducted within the past 5 years and were considered to be representative of the last 20 years of research within this area.

Self-management. Self-management techniques include observation, evaluation, and consequation of one's own behaviour, with the target behaviour and consequences self-selected or specified by somebody else (Carr et al., 2014; Wilkinson, 2008). The essence of self-management is that the young person (to some degree) independently implements behavioural strategies to change their behaviour (Carr, 2016). While standard behavioural interventions rely on external agents (e.g., parents) to modify behavioural contingencies, self-management empowers individuals to learn strategies to regulate their own behaviour (Singh, Lancioni, Manikam et al., 2011).

Self-management procedures of various kinds are established evidence-based procedures for young people with ASD (Carr et al., 2014; Carr, 2016; White et al., 2018), and may be applicable within the current study to enhance participation. Accordingly, self-management techniques have been shown to be particularly effective when applied alongside other evidence-based procedures, such as functional assessment (Wilkinson, 2008). Further, research shows self-monitoring checklists alone facilitate independence in daily care tasks and provide individuals with neurodevelopmental disorders a sense of mastery (Garff & Storey, 1998). However, self-management procedures tend to take longer than other behavioural programmes to produce treatment effects (Singh, Lancioni, Manikam et al., 2011).

A recent meta-analysis of 12 single-subject research studies investigated the effectiveness of self-management to reduce challenging behaviour exhibited by 4- to 18-year-olds with ASD (Carr, 2016). Most studies included discrimination training, whereby the young person was taught to differentiate between appropriate and inappropriate behaviour to facilitate accurate self-monitoring. Nearly all participants were required to either record or monitor their own behaviour, and contingent on accurate reporting, either independently reinforced

themselves or were provided reinforcement from others (Carr, 2016). Results suggest self-management is an effective treatment for reducing challenging behaviour, including inappropriate vocalisations, repetitive behaviour, noncompliance, tantrums, and threats to self-injure, in young people with ASD (Carr, 2016). Interestingly, age was not a predictor of outcomes (Carr, 2016). Accordingly, this research supports the idea that young person-implemented self-management procedures may reduce sleep-interfering behaviours contributing to sleep disturbance.

Few studies have investigated the generalisation and maintenance of self-management techniques and, consequently, the longevity of treatment effects is unclear (Carr, 2016). Additionally, Carr (2016) found only half of the reviewed studies were conducted within a home setting. Despite limited research, Singh, Lancioni, Manikam et al. (2011) suggest treatment durability is probable given individuals can use self-management strategies across settings, and apply the technique to thoughts, feelings, and behaviour. Future research is necessary to investigate the applicability of self-management to sleep disturbance within a home context.

Mindfulness. As discussed previously there is a growing body of evidence suggesting mindfulness techniques can improve insomnia symptoms in typically developing adolescents through reduction of cognitive and physiological arousal (Ong, Ulmer, & Manber, 2012). However, few studies have investigated the efficacy of mindfulness interventions applied to people with ASD (Cachia, Anderson, & Moore, 2016; Hartley, Dorstyn, & Due, 2019; Semple, 2019), and none in the context of a sleep intervention. Cachia and colleagues (2016) and Hartley and colleagues (2019) conducted a systematic review and meta-analysis respectively of mindfulness interventions delivered to individuals with ASD for a diverse range of issues. They identified two studies whereby mindfulness was applied to children (Hwang, Kearney, Klieve, Lang, & Roberts, 2015; Ridderinkhof, de Bruin, Blom, & Bogels, 2018) and four to adolescents (de Bruin, Blom, Smit, van Steensel, & Bogels, 2015; Singh, Lancioni, Manikam et al., 2011; Singh, Lancioni et al., 2011) with ASD. Mindfulness interventions were either implemented via parent-mediated training, concurrently with parents and their child, or directly with the young person (Cachia et al., 2016). Common strategies within mindfulness interventions included contact with the present moment; exploring breathing, mental states and emotions; body scans; as well as development of non-judgemental acceptance of these experiences (Cachia et al., 2016). For example, participants in a study by Singh, Lancioni, Manikam et al. (2011) were taught to refocus their attention from an emotionally arousing event or thought to a neutral part

of the body (the soles of the feet). Individuals were taught how to bring their focus to their body, relax, and then choose how to act (Singh, Lancioni, Manikam et al., 2011).

Overall, the existing literature suggests mindfulness interventions may effectively reduce anxiety, thought problems, and aggression experienced by young people with ASD (Cachia et al., 2016). However, child and adolescent participants experienced smaller gains in wellbeing post-treatment compared to adult participants, regardless of implementation method (Hartley et al., 2019). Additionally, the existing studies in this area lack methodological rigour and are at high risk of publication bias (Cachia et al., 2016; Hartley et al., 2019; Semple, 2019). Most included a small number of participants and lacked measures of treatment fidelity or social validity (Cachia et al., 2016; Semple, 2019). Few investigated the application of mindfulness to children with ASD and all the evaluated studies solely included participants with HFA, therefore the generalisability of mindfulness interventions to a wider population on the autism spectrum is not known (Hartley et al., 2019). Currently, there is insufficient evidence to support the use of mindfulness with children and young people with ASD (Cachia et al., 2016; Hartley et al., 2019; Semple, 2019). Therefore, while mindfulness may be an appropriate technique to facilitate young person involvement, the lack of evidence for this treatment precludes its use within the current study.

CBT. CBT is an efficacious treatment for a range of psychological issues in typically developing children and adolescents (Hofmann, Asnaani, Vonk, Sawyer, & Fang, 2012). Over the past 20 years many studies have examined the effectiveness of modified CBT for young people with ASD (Ho, Stephenson, & Carter, 2018; Perihan et al., 2019). Modified CBT accounts for potential communication and executive functioning differences and involves many of the adaptations outlined in the preceding section. The evidence base for modified CBT has been synthesised in a number of reviews and meta analyses (Burkhart et al., 2018; Donoghue, Stallard, & Kucia, 2011; Ho, Stephenson, & Carter, 2014; Keefer et al., 2018; Kester & Lucyshyn, 2018; Kreslins et al., 2015; Lang et al., 2010; Moree & Davis, 2010; Perihan et al., 2019; Reaven et al., 2009; Rotheram-Fuller & MacMullen, 2011; Scattone & Mong, 2013; Syriopoulou Delli, Polychronopoulou, Kolaitis, Antoniou, & Alexandros-Stamatios, 2018; Ung, Selles, Small, & Storch, 2015; Walters et al., 2016; Weston, Hodgekins, Langdon, 2016; White et al., 2009; Wood, Fujii, & Renno, 2011). Specifically, within the past five years at least nine reviews have examined the evidence base of modified CBT for anxiety in children and adolescents with ASD (Burkhart et al., 2018; Hunsche & Kerns, 2019; Keefer et al., 2018;

Kester & Lucyshyn, 2018; Kreslins et al., 2015; Perihan et al., 2019; Syriopoulou Dellia et al., 2018; Ung et al., 2015; Weston et al., 2016). The following section outlines their findings.

Overall, based on numerous methodologically rigorous studies, modified CBT for anxiety in children and adolescents with HFA can be considered an empirically supported and evidence-based intervention (Burkhart et al., 2018; Ho et al., 2014; Keefer et al., 2018; Kester & Lucyshyn, 2018; Perihan et al., 2019; Walters et al., 2016). Further, findings suggest CBT treatment programmes are superior to alternative control conditions (e.g., another treatment condition; Ung et al., 2015). Promisingly, the moderate to large effect sizes demonstrated within the research coincide with meta-analyses investigating the efficacy of CBT in individuals without ASD (Ung et al., 2015).

Across studies, there was variance in CBT modality (Keefer et al., 2018). Intervention was either delivered to young people by psychologists or therapists in a group or individual format, and parents and/or peers supported participant learning (Ho et al., 2014). The length of intervention varied from 6 weeks to 32 months, with individual sessions taking 1 to 2 hours (Ung et al., 2015). Perihan et al. (2019) found studies which employed standard- (>12 weeks) and long- (> 16 weeks) term CBT interventions had larger effect sizes than short- (< 12 weeks) term programmes (Perihan et al., 2019). Increased intervention sessions likely enabled participants sufficient time to understand and apply therapeutic skills (Ho et al., 2014; Ho et al., 2015; Perihan et al., 2019).

Common components of CBT sessions were psychoeducation, relaxation training, gradual exposure, cognitive restructuring, and problem solving (Kester & Lucyshyn, 2018; Ung et al., 2015; Walters et al., 2016). Modifications to facilitate the learning needs of participants included use of concrete language, visual aids, worksheets, social stories, role-play, and VM (Ho et al., 2015; Ung et al., 2015). Additionally, sessions were highly structured, and participants were often reinforced for engaging in CBT strategies (Kester & Lucyshyn, 2018; Ung et al., 2015).

Behavioural strategies were employed more often than cognitive intervention components (Burkhart et al., 2018; Ho et al., 2015; Walters et al., 2016; Weston et al., 2016). In fact, relaxation and calming behaviours, particularly relevant to sleep disturbance, were the second most frequently addressed skills (Ho et al., 2015). Cognitive restructuring and/or problem-solving techniques were avoided at times due to concerns regarding communication and language abilities, degree of insight, inattention, and low motivation (Ho et al., 2015).

While such strategies typically require higher order cognitive skills and/or extensive verbal ability (Keefer et al., 2018; White et al., 2019), research shows behavioural techniques including gradual exposure, reinforcement, and relaxation, can effectively reduce anxiety in young people with ASD who have low cognitive and verbal abilities (Rosen, Connell, & Kerns, 2016). Cognitively oriented strategies are feasible for children with HFA when cognitive demand corresponds with their cognitive ability and development (Ho et al., 2015). Within the CBT and ASD literature, treatment components for anxiety have not been evaluated independently of one another, so the necessity of specific cognitive and behavioural strategies are unknown (Syriopoulou Delli et al., 2018). Of note, the effectiveness of modified cognitive behavioural interventions for young people with ASD have not been compared to standard cognitive behavioural treatment (Walters et al., 2016). Consequently, it is not possible to attribute the effectiveness of such interventions to the modifications (Walters et al., 2016).

Within the reviewed studies participants were aged 7 to 18 years (Burkhart et al., 2018). The evidence base is particularly strong for children aged 10 to 12 years of age (Ho et al., 2015), however there is insufficient evidence with older adolescents (Keefer et al., 2018). Further, young people with mild to moderate intellectual disabilities were excluded from all studies (Ho et al., 2014; Ho et al., 2015; Ho et al., 2018; Ung et al., 2015; Walters et al., 2016). Consequently, the generalisability of CBT interventions to children and adolescents with less language and cognitive ability is unknown (Kreslins et al., 2015; Walters et al., 2016; Syriopoulou Delli et al., 2018). Although, there is some evidence for the use of CBT with individuals with ID (Keefer et al., 2018). Additional modifications may be necessary to facilitate application to this population (Keefer et al., 2018; Moree & Davis, 2010).

As cognitive behavioural treatment strategies can be utilised to reduce anxiety in children and adolescents with HFA, it is reasonable to expect a combination of these techniques could improve sleep disturbance maintained by anxiety related cognitions and pre-sleep hyperarousal (Paine & Gradisar, 2011; Richdale et al., 2014). Further, as both cognitive and behavioural components can be implicated in the establishment and maintenance of sleep disturbance, interventions which address both areas are likely to be effective in lessening sleep disturbance (Richdale et al., 2014). This is supported by the results of Souders et al. (2017) and McCrae et al's (2019) research which showed parent- and child-implemented cognitive behavioural techniques improved sleep problems in children with HFA. A clear shortfall within the existing literature is the lack of studies investigating individualised, cognitive/behavioural approaches with older children and adolescents diverse in functioning.

Overall, the reviews summarised above have indicated cognitive/behavioural interventions can effectively treat paediatric insomnia in typically developing individuals. Although there are additional challenges when engaging young people with ASD in the therapeutic process, modifications which facilitate assessment and treatment delivery are possible. In addition, the growing evidence base for self-management and modified CBT applied to young people with ASD supports the use of these techniques within the current study. Specifically, it is necessary to assess the applicability of these techniques to treating sleep disturbance, in this population.

Rationale for the Present Research

Untreated, sleep disturbance can become a long-term, lifelong issue, detrimental to multiple aspects of one's life (Cortesi et al., 2010; Goldman et al., 2012; Herrmann, 2016; Hodge et al., 2014; Jin et al., 2012; Richdale & Schreck, 2009; Sivertsen, 2012). More research regarding the treatment of sleep disturbance in young people with ASD is warranted considering the extent of the problem within this population (Moss et al., 2014; Schreck, 2001; Turner & Johnson, 2013; Vriend et al., 2011; Weiskop et al., 2005). Although young people with ASD may present with similar sleep problems often the specific function of the behaviour and contributing environmental factors are unique (Brown & Piazza, 1999; Kodak & Piazza, 2008). Diverse topographies of sleep-related behaviour, in addition to the heterogeneity of autism, necessitate an individualised treatment approach. This can be facilitated by the use of FBA. Based on existing research, it is thought that an FBA-informed intervention will prove more effective than treatments in which the function of the behaviour is ignored. Consequently, the studies in this thesis will use FBA to identify antecedent and consequence variables maintaining sleep disturbance in young people with ASD. Treatments will then be formulated to target these variables.

Most intervention research for paediatric insomnia has employed behavioural or cognitive/behavioural interventions (Meltzer & Mindell, 2014). Such interventions are highly effective at treating sleep disturbance in typically developing young people (Honaker & Meltzer, 2014; Meltzer & Mindell, 2014; Vriend et al., 2011). Further, they lack the side effects of pharmacological treatment and are preferred by parents and health caregivers (Beebe, 2016). However, there has been a distinct lack of methodologically rigorous research studies investigating behavioural and cognitive/behavioural treatments for sleep disturbance in young people with neurodevelopmental disorders (Meltzer & Mindell, 2014). Critically, very few

studies have evaluated sleep interventions for older children (> 8 years) and adolescents with autism (Allik et al., 2006; Turner & Johnson, 2013). Consequently, the efficacy of behavioural or cognitive/behavioural sleep interventions for such individuals is unclear (Richdale & Wiggs, 2005; Turner & Johnson, 2013; Vriend et al., 2011). To address this extant gap in the literature the current research will investigate application of FBA-informed interventions with young people aged 9 to 18 years of age.

Existing literature neglects the inclusion of young people with developmental disabilities as active agents within their own sleep intervention (Strickland-Clark, Campbell, & Dallos, 2000). Interventions whereby young people are actively included within the therapeutic process are not only effective but are also underpinned by a human rights and ethical approach. Further, research shows appropriate modifications (e.g., incorporation of special interests) can be made to standard behavioural interventions to facilitate the involvement of young people with ASD (Attwood & Scarpa, 2013; Moree & Davis, 2010; Reaven, 2009; Walters et al., 2016; Wood & Schwartzman, 2013). Additionally, there is a strong relationship between child and youth participation in therapy and successful treatment outcomes (Karver, Handelsman, Fields, & Bickman, 2006). Involving adolescents directly in the therapeutic process of sleep enables them to increase their knowledge about appropriate sleep behaviour and coping mechanisms and teaches them the skills to resolve their sleep disturbance (Schlarb et al., 2011). Finally, a core aspect of development in adolescence is individuation and separation from parents; while parents can play an important role within sleep interventions by encouraging and enforcing sleep-conducive behaviour, it is essential older children take on a more active role (Clarke et al., 2015; Vriend & Corkum, 2011). Consequently, both young person- and parent-implemented treatment components will be employed within the current research, rather than parents or practitioners acting as the sole agents of change. These terms are differentiated by who is primarily responsible for implementing the component (e.g., relaxation training is young person-implemented, external reinforcement of sleep-conducive behaviour is parent-implemented) and/or who it is primarily directed to (e.g., psychoeducation could be directed to both the young person and their parents).

Although cognitive (e.g., ‘detective thinking’) and behavioural (e.g., PMR) treatment strategies for sleep disturbance can be engaged in by typically developing children and adolescents, the feasibility of these methods for use by young people with ASD needs further investigation. For example, features of ASD and impaired communicative and/or cognitive abilities may limit young people’s capacity to understand and apply FBA-informed intervention

components with high procedural integrity, even if appropriate modifications are carried out. Additionally, the capacity for young people with ASD to take primary responsibility for maintaining healthy sleep practices (e.g., selecting and implementing a relaxing bedtime routine), without any parent input, has not been established. Consequently, these factors will be explored in the current thesis.

Young person-implemented intervention components (e.g., psychoeducation, relaxation instruction, protective items) may reduce reliance on traditional parent-implemented behavioural sleep interventions (e.g., unmodified extinction). In accordance with the principles of minimal sufficiency and the least restrictive alternative, it is ethical practice for treatment to consist of the least time intensive, intrusive, and aversive methods capable of producing significant therapeutic change (Kazdin, 1984; Sanders, Kirby, Tellegen, & Day, 2014). Although there are no specific guidelines outlining the least to most restrictive applied behaviour analytic interventions, antecedent-based interventions and reinforcement are generally perceived to be less aversive or drastic than punishment techniques (e.g., extinction; Bailey & Burch, 2013; Kazdin, 2013). Within the present research the following least to most restrictive ranking system will be applied: antecedent-based interventions (e.g., sleep hygiene, consistent bedtime routine, social story, VSM), positive reinforcement (e.g., reward system), modified extinction (e.g., systematic fading of parental presence), unmodified extinction (e.g., removal of all electronic devices and/or toys from the bedroom), negative punishment (e.g., response-cost), and positive punishment (e.g., reprimand). The present research aimed to apply the least restrictive and minimally sufficient interventions possible and evaluate whether ASD-related sleep disturbance can be addressed via such methods.

In general, young people with ASD and lower intellectual abilities have tended to be excluded from studies involving young person-implemented components (e.g., CBT). The three studies which incorporated young person-implemented components in cognitive/behavioural sleep interventions for young people with ASD only included participants with HFA (Loring et al., 2016; McCrae et al., 2019; Souders et al., 2017). Within the current thesis, inclusion criterion will purposefully not be limited by IQ. Instead, participants will include young people with sufficient communication abilities to engage in the therapeutic process (hereafter such young people will be referred to as verbal individuals). This ensures young person-implemented components are evaluated with children and adolescents with diverse levels of functioning.

Within the typically developing paediatric sleep literature, the value of the young person's perspective of the sleep problem/s is recognised, and they are included within the assessment and treatment evaluation process. Semi-structured interviews, self-reported sleep diaries, and psychometrics have been used to obtain information directly from participants for which caregiver report cannot necessarily be relied (Hendricks et al., 2014; Norell-Clarke, Nyander, & Jansson- Fröjmark, 2011; Paine & Gradisar, 2011; Stewart & Gordon, 2014). Specifically, these are the intensity and content of sleep related cognitions, beliefs, and attitudes; covert sleep-interfering behaviour; sleep quality; and young person ratings of treatment acceptability (Lewis et al., 2015; Rafihi-Ferreira et al., 2018; Stewart & Gordon, 2014). Within this thesis a combination of parent- and self-report (e.g., sleep diaries, questionnaires) measures, as well as VSG will be utilised to collect such information. Actigraphy will not be used for a variety of reasons. Firstly, the device must be regularly retrieved from families in order to download and process data, therefore it cannot provide continuous data over an extended time period (Moore et al., 2017). Secondly, young people with ASD may not tolerate device placement on their person (Katz, Malow, & Reynolds, 2016; Moore et al., 2017). Finally, it cannot reliably identify inactive wake periods or capture salient information regarding topographies of sleep and awake behaviours, unlike VSG (Katz et al., 2016; Moore et al., 2017).

Social validity is a crucial component when evaluating intervention effectiveness; the methods employed should be acceptable to all consumers and produce meaningful, socially important outcomes (Callahan et al., 2017; Kazdin, 2000; Wolf, 1978). Although behavioural sleep interventions are generally accepted by parents (Beebe, 2016), no existing study in the autism and sleep literature has assessed young peoples' perspectives. Given children and adolescents directly experience the interventions as well sleep disturbance itself, arguably their opinion is of utmost importance.

Critically, many adverse effects of sleep disturbance have the propensity to be reversed if sleep issues can be ameliorated, emphasising the importance of changing sleep to improve the overall functioning of young people and their families (Richdale & Wiggs, 2005). Despite numerous studies emphasising the association between sleep disturbance and detrimental wellbeing, few studies have investigated the impact of sleep interventions on the mental health and functioning of the child and/or their caregivers. Consequently, more research is needed which evaluates potential secondary outcomes of sleep interventions for young people with ASD.

Research Questions

The present research aimed to answer the following three questions in part via investigation of each sub-question:

1. Are FBA-informed interventions which include young person- and parent-implemented treatment components effective at reducing sleep disturbance in verbal young people with ASD aged 9 to 18 years?
    a. Are treatment effects maintained in the short- and long-term? (Study 1, 2, and 3)
    b. Are young person-implemented interventions feasible for use by verbal 9- to 18-year-olds with ASD (i.e., do the young people implement the treatment plan with high procedural integrity/fidelity? Can they effectively engage in young person-implemented treatment strategies, such as PMR)? (Study 1)
    c. To what extent are parent-implemented intervention components necessary to reduce sleep disturbance? Can young person-implemented treatment components alone effectively reduce sleep disturbance? (Study 2)
    d. Can minimally sufficient and least restrictive FBA-informed intervention approaches (e.g., antecedent-based intervention) address sleep disturbance in a verbal young person with ASD? (Study 3)
2. Are the selected intervention approaches acceptable to participants and their parents across Studies 1, 2, and 3?
3. Will there be changes in participant and/or parent wellbeing following the implementation of selected sleep interventions employed in Studies 1, 2, and 3?
    a. Will there be changes to the following participant wellbeing variables?
        i. Externalising behaviour?
        ii. Internalising behaviour?
        iii. General challenging behaviour (e.g., attention/social/thought problems)?
        iv. Anxiety?
        v. Autism symptom severity?

b. Will there be changes to the following parent wellbeing variables?

    i. Depressive symptoms?

    ii. Anxiety symptoms?

    iii. Stress symptoms?

    iv. Sleep problems?

    v. Marital relationship quality?

Chapter 3

Overview of the Present Research

Purpose

This thesis presents three empirical studies intended to address the gaps in the literature discussed in Chapter 2. Overall, the purpose of these studies was to evaluate the efficacy of FBA-informed sleep interventions designed to include verbally communicative older children and adolescents with ASD in the therapeutic process. The first study (Chapter 4) was a pilot study designed to investigate the feasibility, effectiveness, and acceptability of behavioural sleep interventions including input from the young person with ASD. The second study (Chapter 5) aimed to extend the findings of Study 1 with more cases (providing more opportunities for replication) and with as much participant input as feasible, only including parent-implemented treatment components when necessary. The third study (Chapter 6) employed FBA-informed young person- and parent-implemented components sequentially to evaluate the active treatment components and ensure intervention was least restrictive and minimally sufficient. Finally, in Chapter 7, parent and self-report sleep outcomes (according to questionnaires) are presented and the secondary outcomes of the sleep interventions are evaluated.

The Sleep Research Team

The current research is part of a wider research project investigating ASD and sleep. This project is led by three senior academics (two registered clinical psychologists and an Applied Behaviour Analysis practitioner), assisted by a team comprising registered psychologists, intern psychologists, post-graduate students, and research assistants. The author was an intern psychologist and was responsible for conducting and managing all clinical assessment and intervention procedures with families, with assistance from the wider team. These responsibilities were carried out under the supervision of the aforementioned academics.

Methodology

Nomothetic research (intended to understand the general laws of a group of individuals) has dominated the field of psychology (Blampied, 2013b). Traditional between-group research, involving large samples, random assignment, and null hypothesis statistical testing, is considered the ‘gold standard’ method to establish treatment effectiveness (Kazdin, 2011). Such techniques may minimise threats to internal and external validity and enable researchers to draw

inferences regarding intervention effectiveness. However, careful control of conditions in between-group research designs (e.g., strict inclusion criteria, a manualised intervention approach applied to all participants) may actually reduce generalisation to clinical or everyday settings where experimental conditions cannot be so meticulously regulated and there is likely to be more complexity (e.g., comorbid disorders; Kazdin, 2011). Further, calculation of aggregate-group data in therapy outcome research, conceals individual results (Blampied, 2013b; 2017) and the average may not apply to any individual clinical case (Barlow & Nock, 2009). Conversely, idiographic approaches (the study of one individual case) enable in-depth analysis of clinical cases (Blampied, 2017). Similar to between-group research designs, single-case research designs can also enable researchers to draw inferences regarding the causal relation between variables (Barlow & Nock, 2009). Empirical single-case designs include a continuous baseline phase to establish the trajectory of the dependent variable in terms of its variability, level, and trend. This projects the case's future performance without intervention, thus baseline acts as a within-case control condition. Inferences about intervention effectiveness are supported when improvement in target behaviour coincides with the commencement of intervention and is maintained over time (Kazdin, 2011). Replication across participants, behaviours, and settings reduces threats to external and internal validity (Cohen, Feinstein, Masuda, & Vowles, 2014) and is the basis for making inferences regarding treatment effects.

Given the heterogeneity of young people on the autism spectrum, single-case research with such individuals is highly appropriate (Carr, 2016). A primary objective of the current research was to create individualised interventions for each participant based on idiosyncratic FBA results. Therefore, a between-group research design whereby aggregate-participant data is used to evaluate treatment outcomes would not have been suitable. Single-case designs enable researchers to gather substantial, personalised data sets (Kazdin, 2011). Further, individualised interventions can be adapted accordingly based on observed target behaviour (Lane & Gast, 2014). For these reasons, single-case designs are well suited to preliminary research, especially when the investigation involves therapy innovations, as in the current research (Blampied, 2013b; Dubois & Gadde, 2002; Kazdin, 2011).

General Method

Experimental Design

Each study in the current thesis employed a non-concurrent (i.e., cases entered treatment at different times) single-case design with random allocation of baseline lengths to evaluate the

effectiveness of FBA-informed sleep interventions. Study 1 utilised an AB single-case design with replication of treatment effects across target behaviours and participants (N = 3). AB single-case studies refer to the application of two adjacent conditions (baseline [A], intervention [B]) with a specific set of variables under which the target behaviour is measured (Lane & Gast, 2014). Study 2 employed a multiple-baseline design across participants and behaviours (N = 8). Multiple-baseline designs allow for repeated demonstrations of behaviour change following staggered implementation of intervention conditions, thus reducing the likelihood subsequent treatment effects were influenced by extraneous factors (Kazdin, 2001). Study 3 employed an AB single-subject design with replication of treatment effects across target sleep variables (e.g., CCs, SOL, NWs, total sleep duration, and SE). Intervention consisted of three sub-phases (B1, B2, B3) with cumulative addition of treatment elements, to establish active components and ensure treatment was minimally sufficient.

The addition of a reversal phase was considered for Studies 1 and 3 to reduce threats to validity. This ABAB design can provide additional evidence that behaviour change is attributable to intervention when behaviour improves during the first intervention phase, reverts to previous baseline levels when treatment is withdrawn, and improves once again when intervention is reinstated (Kazdin, 2011). This did not seem appropriate in the current study as in some cases, intervention may lead to the acquisition of new skills for young people and their parents and thus a return to baseline would not have been possible. Further, as sleep disturbance is often detrimental to both an individual and their family's wellbeing, the removal of an intervention designed to address such issues would have been unethical. Finally, as individuals with ASD often prefer consistency and structure within their daily lives, frequent changes between experimental phases may have been distressing/disruptive and unnecessarily aversive for participants.

Data Analysis

Intervention was deemed to be effective if a functional relation between target behaviour and treatment was demonstrated, whereby systematic changes in behaviour were clearly related to the introduction or alteration of treatment conditions (Kazdin, 2001). Target behaviour was measured daily over an extended time period to analyse patterns and stability (Kazdin, 2011). Treatment effects were judged based on changes from the previously projected performance (Kazdin, 2011). Continuous assessment, as opposed to pre and post measurement, facilitated the

prediction of future target behaviour and thus increased the internal validity of the research findings (Kazdin, 2011).

Visual analysis of graphed target behaviour was used to assess treatment effectiveness. In accordance with guidelines specified by Lane and Gast (2014), visual analysis included assessment of change in the trend, level, and stability of data. Trend describes the direction or progression of the data pattern; level can be defined as the location of the data points relative to their possible minimum and maximum; and stability is the consistency of data magnitude across a condition, or lack of variability (Lane & Gast, 2014). Stable baseline data strengthens the inference that change which co-occurs with treatment is attributable to the intervention itself (Kazdin, 2011). When no trend is apparent within baseline, or is in a non-therapeutic direction, therapeutic change upon treatment implementation provides evidence for its effectiveness (Kazdin 2011). However, if the baseline trend suggests participant behaviour is already improving, further therapeutic change during the intervention phase cannot necessarily be attributed to treatment (Kazdin, 2011). Changes in data level indicate whether behaviour is improving, worsening, or merely plateauing across intervention phases (Lane & Gast, 2014) If data in baseline is consistently close to maximum or minimum levels this may represent a ceiling or floor effect and impair detection of any subsequent change.

Visual analysis was supplemented with descriptive statistics to strengthen the conclusions drawn regarding treatment effects. Effect size calculations for group design research, such as Cohen's *d*, are not suitable for single-case studies, as they tend to inflate scores (Roth, Gillis, & DiGennaro Reed, 2014). One of the most popular effect size metrics developed for single-case research includes nonoverlapping methods (Roth et al., 2014). Nonoverlapping methods calculate the extent to which intervention and follow-up data do not overlap with baseline data, thus indicating behaviour change (Parker & Vannest, 2009). The higher the percentage of nonoverlapping data, the greater the behaviour change between baseline and intervention or follow-up phases. Percentage exceeding/below the median (PEM/PBM) combines nonoverlap methods with median level change (Parker, Vannest, & Davis, 2011). This technique compares the percentage of intervention data points below or exceeding the baseline median (depending on the therapeutic direction of change) to evaluate the degree of behaviour change. For example, if the baseline median was 100 and 25/25, intervention data points were below 100 (PBM = 100%), whereby if 50% of the data points were below 100 (PBM = 50%). PEM/PBM interpretation is as follows: < 70% represents ineffective treatment; 70 to 90% moderate effectiveness; and > 90% high effectiveness (Ma, 2009). PEM/PBM assumes the

baseline median is an accurate representation of data within this phase (Parker et al., 2011). Although median level change statistics are superior to mean level change statistics, accurate interpretation is still compromised when data sets in a phase are highly variable and lack central tendency (Parker et al., 2011). Further, PEM/PBM is insensitive to the absolute magnitude of data points, therefore, a score of 100% could be obtained if all treatment data points were only slightly different to the baseline median (Ma, 2006). However, the use of PEM/PBM in conjunction with visual analysis, alleviates these concerns; visual analysis guidelines would conclude there had been little change if the level reduction was only slight, despite all intervention data points being below or above the baseline median.

Modified Brinley plots were utilised to evaluate change in psychometric scores from pre- to post-treatment at the idiographic and nomothetic level (Blampied, 2017). A modified Brinley plot is a scatterplot which depicts the values of a single dependent variable at two time points (Blampied, 2017). The reliable change index (RCI) was used to ascertain whether differences in pre and post primary and secondary outcome data were reliable; i.e., larger than that likely due to measurement error alone (Jacobson & Truax, 1991). RCI provides a conservative assessment of objective change as a result of treatment, by accounting for test variability and measurement error (de Souza Costa & de Paula, 2015). If the change in scores from pre- to post-test are in excess of what could be expected from measurement error alone, the change is considered reliable. This is assessed by calculating the standardised change score (dividing the change in pre- and post-test psychometric scores by the standard error of the difference) and comparing it to a criterion of +/-1.96. Scores exceeding +/-1.96 are indicative of reliable change ($p < .05$). The standardised change score (SCS) was calculated (see below) using Equation 1 (where S_{Diff} = standard error of difference; *SEM* = standard error of measurement; *SD* = normative sample standard deviation; and r_{xx} = Chronbach's α or test-retest reliability).

$$\text{SCS} = \frac{x^2 - x^1}{SDiff}$$

$$S_{Diff} = \sqrt{2(SEM)^2} \quad (1)$$

$$\text{SEM} = SD\sqrt{1 - rxx}$$

The RCI is the minimum raw score difference required for the change to be reliable, calculated by multiplying the standard error of difference by 1.96 ($S_{diff} \times 1.96$).

Clinical significance. Importantly, the reliability of change and the size of a treatment effect do not convey whether improvement was clinically or socially significant. In the current study the clinical significance of secondary outcomes was evaluated for all participants, and the clinical significance of sleep outcomes was formally evaluated in Study 3. Clinically significant change was defined as elimination of the presenting problem, evidence of typical functioning, or a significant reduction in the likelihood of problems (e.g., target behaviour changes from clinical to normative levels; Jacobson, Follette, & Revenstorf, 1984). It is likely that clinical change will be underestimated in this thesis as normative data was mostly based on non-clinical populations, whereby behaviour may be less resistant to change. Further, clinically significant change may not be an appropriate indication of meaningful treatment effects when a return to normative or standard levels of functioning would not be expected (Kazdin, 1977; Wise, 2004). Accordingly, in the current research we would not expect an individual with a neurodevelopmental disorder to 'recover' or show no indication of clinical problems, but instead perhaps improve sufficiently to benefit their independent functioning or to a more desirable/manageable level.

Procedure

Each study consisted of assessment (including FBA), baseline, intervention, and follow-up phases. A comprehensive FBA based on clinical interviews, the SATT, Questions About Behavioral Function (QABF; Matson & Vollmer, 1995), sleep diaries, and analysis of video footage, was conducted to inform intervention design. Baseline began following FBA and served to provide a basis for subsequent treatment comparison. Families were asked to continue their typical sleep practices during baseline to ensure existing behaviour patterns were accurately obtained.

FBA-informed sleep interventions commenced immediately once the randomly assigned baseline period ended. Intervention consisted of emerging, evidence-based, cognitive and behavioural treatment components, implemented by the young person and parent (s) as necessary. An analysis of young person- versus parent-implemented intervention conditions was considered, however, at times FBA results necessitated a combined approach from the outset. For example, parent-delivered reinforcement was deemed necessary to encourage participants to engage in desired behaviour (e.g., remain in their bed post-bedtime) when they were not motivated to do so and particularly when it was distressing for them. Specific intervention components applied with each family are detailed within the individual studies. The intervention

phase continued until there was a significant reduction in sleep disturbance evidenced over a minimum 14-day period.

Short and long-term follow-up were typically conducted between 3 and 20 weeks post-treatment respectively, allowing for assessment of the maintenance of behaviour change over time. A secondary collection of long-term follow-up was able to be undertaken in Study 1 at 18 to 24 months post-treatment

Participants

Ethics. Ethical approval for this study was provided by the University of Canterbury Human Ethics Committee (HEC 2018/47; see Appendix A).

Recruitment. Families were recruited through flyers (see Appendix B) shared to autism community groups on social media and also provided to NZ organisations and service providers for young people with ASD and their families. Participants were also referred by professionals within these organisations.

Eligibility. Participants were included if they met the following eligibility criteria (a) a formal diagnosis or features of ASD as verified by a psychiatrist, psychologist, or paediatrician, and supported by results of the Gilliam Autism Rating Scale - Third Edition (GARS-3, Gilliam 2013); (b) aged between 9 and 18 years; (c) parent-reported unwanted co-sleeping or difficulty initiating and maintaining sleep, supported by systematic in-home measurement; (d) no medical condition that directly interfered with sleep; and (e) sufficient receptive and expressive communication skills to engage in the assessment and treatment processes. The Communication criterion was assessed through clinical judgment coupled with responses to items on the Communication sub-domain of the Vineland Adaptive Behavior Scales Second (VABS-II; Sparrow, Cicchetti, & Balla, 2005) or Third Edition when the most recent version was published in 2016 (Vineland-3; Sparrow, Cicchetti, & Saulnier, 2016). For example, the Vineland items considered included, “Follows directions to do something with one object” and “Says at least 50 words”.

Characteristics of participants. Fifteen participants met eligibility criteria. One participant withdrew following the assessment phase and two participants completed baseline data and withdrew prior to beginning intervention. Reasons cited for withdrawing included improvement in sleep or external stressors preventing involvement. The remaining participants

included 11 males and 1 female aged between 9 and 15 years. Most participants' ethnicities were identified by their parent/s as being NZ/European, with one participant identifying as NZ/Māori. Three participants were of English, South African, and Canadian nationalities respectively. Parent reported sleep disturbance included, CCs, delayed SOL, NWs, EWs, and unwanted co-sleeping. Six participants regularly took melatonin; however, this was not sufficient to resolve their sleep difficulties. Participant communication abilities were generally lower than same age/same gendered peers, and receptive abilities tended to be less developed than expressive abilities. Many participants had diverse comorbid psychiatric conditions, including ID, ADHD, and generalised anxiety disorder (GAD). The families represented a range of socioeconomic status, as indicated by the *New Zealand Socio-economic Index 2013* (NZSEI-13; Fahy, Lee, & Milne, 2013). Additionally, eight participants resided in two-parent homes and four participants resided in one-parent households. Characteristics of each participant and their families are detailed in Table 3.1.

Setting

The research team was based in Christchurch, NZ; however, participants included families who lived throughout the country. Eligibility criteria were evaluated over the phone. Information sheets and consent forms were subsequently mailed to eligible participants (Appendices C, D, E, F, G, H, I, and J). Clinical interviews, treatment planning, and progress discussions were conducted at the Pukemanu Centre at the University of Canterbury, at participants homes, or via Skype. Treatment was implemented within the home by participants and their parents with remote support from the authoring intern psychologist. Pre- and post-treatment questionnaires were posted to families or completed over the phone.

Measurement

Measurement of primary and secondary outcome variables was conducted using VSG, clinical interviews [Appendices K, L, T, and U], parent- and self-report sleep diaries [Appendices M and N], and psychometric measures.

Table 3.1. *Summary of Participant Characteristics in Studies 1, 2, and 3 at commencement of intervention*

Participants	Age (Y-M)	Sex	Nationality	Diagnosis	Medication	Education	SES	Parents Residing	Receptive/ Expressive/ Written Communication Age Equivalent	Sleep Disturbance
Study One										
Niko	9-7	Male	NZ/Euro	Asperger's	-	Mainstream school (teacher aide support)	24	1	2-5 4-11 6-1	NWs EWs
Peter	14-6	Male	English	ASD	Melatonin Risperidone Fluoxetine	Specialist school	90	2	2-10 3-11 7-0	Delayed SOL EWs
Eric	11-6	Male	NZ/Euro	ASD	-	Mainstream school	50	1	5-6 12-3 10-8	CCs Delayed SOL
Study Two										
Blair	9-0	Male	NZ/Euro	ASD Chromosomal duplication 15q13.3	Atomoxetine Risperidone	Home school	37	2	1-3 3-1 4-3	NWs
Seth	9-6	Male	NZ/Euro	ASD ADHD TS APD	-	Mainstream school	77	2	2-10 10-6 11-9	CCs Delayed SOL
Will	11-6	Male	NZ/Euro	ASD	-	Mainstream school	37	2	4-0 4-8 10-0	Unwanted co-sleeping
Finn	11-11	Male	NZ/Euro	Features of ASD ADHD Dyspraxia	Melatonin	Mainstream school	72	2	7-3 4-2 8-4	CCs
Ben	12-2	Male	South African	ASD GAD	Melatonin	Mainstream school	56	2	3-5 5-10 7-6	CCs NWs

Scott	12-4	Male	NZ/Euro	ASD ID Dyspraxia	Melatonin Fluoxetine	Mainstream school (teacher aide support)	48	2	1-7 3-2 7-0	Unwanted co-sleeping
John	14-2	Male	NZ/Euro	ASD ADHD TS Dyslexia Irlen Syndrome APD	Melatonin Clonidine Atomoxetine	Mainstream school	72	1	4-4 5-6 7-0	CCs Delayed SOL
Isaac	15-7	Male	NZ/ Māori	ASD	-	Mainstream school	44	1	22-0 21-0 20-0	Delayed SOL
Study Three										
Eve	9-9	Female	Canadian	ASD Selective mutism	Melatonin	Part-time mainstream school and home school	68	2	2-9 3-11 8-6	CCs Delayed SOL NWs

Note. SES = Socioeconomic status; NZ/ Euro = New Zealand/ European; ASD = autism spectrum disorder; ADHD = attention-deficit/hyperactivity disorder; APD = auditory processing disorder; GAD = generalised anxiety disorder; TS = Tourette's Syndrome; ID = intellectual disability; NWs = night wakings; EWs = early wakings; SOL = sleep onset latency; CCs = curtain calls. SES was assessed by the New Zealand Socio-economic Index 2013 (Fahy et al., 2013), based on the occupation of the principal income earner. Ten = lowest SES and Ninety = highest SES. Age equivalent communicative abilities were assessed using the Vineland Adaptive Behavior Scales Second and Third Edition, Caregiver Rating Form (Sparrow et al. 2005; Sparrow et al., 2016).

Chapter 4: Study 1

Behavioural Sleep Intervention for Young People with ASD: a Pilot Study[1]

Difficulties initiating and maintaining sleep as well as other topographies of sleep disturbance are a common clinical problem for individuals with ASD and their families. In a study of 1,518 children with ASD, Malow, Katz et al. (2016) found that 71% had clinically significant sleep problems, a much higher rate than in samples of typically developing individuals (e.g., Elrod et al., 2016). Although the exact cause of sleep problems in people with ASD likely varies across individuals, previous research suggests their sleep is impacted by a complex interaction between physiological (e.g., dysregulated melatonin), environmental, and behavioural (e.g., inadvertent reinforcement of sleep-interfering behaviour) variables (Richdale and Schreck, 2009). A range of sleep problems have been identified in people with ASD including delayed SOL, reduced total sleep time, reduced SE, and daytime fatigue (Baker, Richdale, Short, & Gradisar, 2013). These issues appear to be particularly problematic during adolescent years (Baker et al., 2013; Goldman et al., 2012). This is consistent with findings involving typically developing adolescents wherein physiological vulnerabilities (e.g., changes in circadian phases), environmental factors (e.g., increased internet use, responsibilities at school and work), schedule changes (e.g., early school start times), and increased autonomy (e.g., freedom to choose their own bedtime and bedtime routine) appear to increase risk for sleep disturbance (Loring et al., 2016). These risk factors can be exacerbated among individuals with ASD whereby deficits in executive functioning may inhibit the organisation and regulation of sleep-conducive bedtime behaviour (e.g., following a consistent bedtime routine, limiting caffeine consumption, restricting screen use at night; Quist, Chaplin, & Hendey, 2015). In addition, preliminary research suggests adolescents and young adults with ASD are more likely to have dysregulated levels of melatonin compared with typically developing controls (Tordjman et al., 2012).

Clinically significant sleep problems are detrimental to the overall wellbeing of individuals and their families. Specifically, insufficient sleep has been linked to increased severity of autism symptomatology, challenging behaviour, and poor psychological wellbeing, as well as compromising parental sleep, quality of life, and marital relationships (Cortesi et

[1] An article based on this study has been published in Advances in Neurodevelopmental Disorders: van Deurs, J. R., McLay, L. K., France, K. G., Blampied, N. M., Lang, R. B., & Hunter, J. E. (2019). Behavioral sleep intervention for adolescents with autism spectrum disorder: a Pilot study. *Advances in Neurodevelopmental Disorders, 3,* 397–410. doi:10.1007/s41252-019-00123-z (see Appendix V).

al., 2010; Herrmann, 2016). Without effective intervention, sleep problems experienced by young people with ASD may persist over time, compromising future functioning across home, school, and work settings.

Behavioural interventions have been effective in treating sleep disturbance in children with ASD (Cuomo et al., 2017). Increasingly, these interventions are informed by FBA, to identify the variables influencing sleep problems and inform selection of individualised, multicomponent, function-based treatments (e.g., Jin et al., 2013; McLay, France, Blampied et al., 2019; McLay, France, Knight et al., 2019). Although there is evidence to support the use of function-based treatments for sleep problems in young children with ASD, there is little research on the utility of FBA-informed sleep interventions for older children and adolescents with ASD. Instead, pharmacological approaches, in particular melatonin, have been the most thoroughly researched treatment (Cuomo et al., 2017).

Most existing sleep-intervention research with typically developing young people involves the young person as the primary change agent. Specifically, involving young people in the sleep intervention increases their knowledge of sleep-conducive behaviour and teaches them skills to resolve their own sleep disturbance (Schlarb et al., 2011). A systematic search of the autism and sleep literature in August 2018, revealed only one study had actively included young people with ASD over 9 years of age in the therapeutic process. Loring et al. (2016) provided two sleep-education sessions to 18 adolescents (11 to 18 years old) with HFA and their parents. Session one targeted sleep hygiene including bedtime routine and arranging the sleep environment, and session two taught relaxation and distraction techniques to facilitate sleep onset. Subsequent application of these practices by the adolescents, with parental support, resulted in significant improvements in sleep onset and efficiency.

When modifications are made to standard therapies to facilitate the engagement of young people with ASD, it is important to assess the social validity and acceptability of the procedures and include input from the young person (Callahan, Shukla-Mehta, Magee, & Wie, 2010). Unfortunately, most social validity data have been collected via parent report, even though parents did not directly experience the targeted sleep disturbance or treatment. Further, reliability between parent-reported sleep diaries and objective sleep measures (e.g., VSG) has predominantly been assessed with younger pre-adolescent children. Parents may be less likely to detect an adolescent's covert sleep-interfering behaviour (e.g., electronic device use), and the validity of parent-reported sleep outcomes should not be assumed.

A number of questions remain about the engagement of young people with ASD and sleep disturbance in their own treatment. First, the feasibility of including young people with ASD, across a range of intellectual functioning in the assessment and treatment process is not well established. For example, it is not known whether young people with ASD are able to complete intervention components effectively and with fidelity. Second, the long-term effectiveness of behavioural sleep interventions for young people with ASD is understudied; specifically, there appears to be no study with follow-up beyond 12 months (Durand, 2002; Weiskop et al., 2001). Third, little is known regarding young person perspectives and social validity of behavioural sleep interventions, which would seem particularly important when interventions include components delivered directly to the participant. Finally, although parent-reported sleep diaries are considered reliable among pre-school-aged children (Hodge, Parnell, Hoffman, & Sweeney, 2012), the reliability of parent-reported sleep in older children and adolescents with ASD warrants consideration. The present study evaluates (a) the feasibility and effectiveness of individualised behavioural sleep interventions involving input from young people with ASD; (b) long-term maintenance of effects; (c) social validity of treatment components implemented with young people for sleep disturbance; and (d) the reliability and validity of parent-reported sleep diaries for young people with ASD.

Method

Participants

Participants ranged in age from 9 to 14 years old and were referred to the research study by their parents or by professionals delivering services to individuals with ASD and their families. Each participant met the following inclusion criteria: (a) formal diagnosis of ASD or Asperger's syndrome, as verified by a psychiatrist, registered psychologist, or paediatrician, and supported by a high probability estimate (Very Likely) and severity level rating on the GARS-3; (b) parent-reported difficulty initiating and maintaining sleep, supported by systematic in-home measurement; (c) no medical condition that directly interfered with sleep; and (d) sufficient receptive and expressive communication skills to engage in treatment. Participant characteristics are summarised in Table 4.1.

Procedures

Design. A single-case design, incorporating baseline [A], intervention [B], and short- and long-term follow-up phases, was used to evaluate treatment effects. Additional intervention

phases are indicated by a phase-change line and alphabetisation (C, D, or E) on Figures 4.3 to 4.5. An AB design was applied to Niko and Eric, and an ABCDE design to Peter.

Table 4.1. *A Summary of Participant Characteristics at Commencement of Intervention*

Characteristics	Niko	Peter	Eric
Age (Y-M)	9-7	14-6	11-6
Sex	Male	Male	Male
Diagnosis	Asperger's Syndrome	ASD	ASD
VABS-II Receptive & expressive language age equivalent (Y-M)	2-5 4-11	2-10 3-11	5-6 12-3
Educational environment	Mainstream school (teacher aide support)	Specialist school	Mainstream school
GARS-3	118 Very likely	108 Very likely	106 Very likely
Medication	_	Melatonin (3mg) Risperidone (0.25–0.5mg) Fluoxetine (20mg)	_

Note. VABS-II = Vineland Adaptive Behavior Scales, Second Edition; GARS-3 = Gilliam Autism Rating Scale-Third Edition.

Setting. Participants were located throughout NZ. Clinical interviews and treatment planning discussions with families were conducted in a university-based clinic or at the participant's home if they were unable to travel to the clinic. The VABS-II and GARS-3 were administered to caregivers over the phone prior to the clinical interview. Other pre-treatment psychometric assessments were given to the parent at the clinical interview to complete and return to the researcher. Post-treatment questionnaires were sent to families upon conclusion of the intervention phase. Treatment was implemented within the participant's home by young people and their parents with the support of the researcher. During treatment, communication with participants and families was conducted in person or via Skype, telephone, and email contact, depending on geographical location and participant preference.

FBA. A combination of the SATT, QABF, sleep diaries, and analysis of video footage was used to conduct the FBA. The SATT is an open-ended interview tool designed to identify specific sleep problems and the unique antecedent and consequence factors which may underlie them. It was used to guide questions in the clinical interview. The QABF, a brief functional assessment checklist used to establish the primary function of sleep-interfering behaviour, was completed by parents following the clinical interview. Sleep diary data and video footage were collected for at least one week during the assessment phase. Information gathered from these sources about the history and type of sleep problems, sleep hygiene practices, antecedent and consequence variables maintaining the sleep problem, and its possible function was synthesised in an FBA-informed case conceptualisation (Blampied, 2013a). In accordance with the bioecological model of human development (Bronfenbrenner, 1979), critical systemic and dynamic factors (e.g., parent-child relationship) were identified in the case formulation and addressed in the subsequent treatment plan if necessary.

Baseline. Baseline commenced following completion of the FBA. Baseline length was staggered such that Peter, Niko, and Eric completed 3, 5, and 8 weeks of baseline recording respectively. Baseline length was determined by random assignment, though on occasion this was extended due to participant readiness to commence intervention (i.e., to ensure that the conclusion of baseline was commensurate with the beginning of treatment, baseline was occasionally extended). During baseline, families were asked to continue with their existing sleep practices (e.g., bedtime routine, electronic device use).

Intervention. Individualised, FBA-based, multicomponent interventions commenced upon conclusion of baseline. The chosen treatment was discussed with parents and participants using the guided participation model to ensure a shared understanding of the issues and intervention (Sanders & Burke, 2014). Treatment included components implemented with both parent and participant. Each treatment component was selected to address hypothesised factors underlying participant sleep disturbance, facilitate treatment compliance, and support the maintenance of helpful sleep habits. Table 4.2 includes a summary of each participant's sleep problem, FBA data, and subsequent intervention components.

Table 4.2. *Problem Behaviour, Factors Precipitating and/or Maintaining Behaviour, Hypothesised Function, and Parent and Young Person Treatment Components for All Three Participants*

	Niko		Peter		Eric	
	Frequent and prolonged NWs	Frequent EWs	Delayed SOL	Frequent EWs	Frequent CCs	Delayed SOL
Factors thought to be precipitating and/or maintaining behaviour	Lack of physiological sleep pressure; electronic device use; adolescent-reported discomfort in bed; warm and comfortable sleep-interfering environment	Lack of physiological sleep pressure; electronic device use; adolescent-reported discomfort in bed; warm and comfortable sleep-interfering environment	Lack of physiological sleep pressure; inappropriate sleep dependencies (electronic device, bright light); lack of discriminative stimuli for sleep; electronic device use; exposure to bright nightlight	Lack of physiological sleep pressure; inappropriate sleep dependencies (electronic device, bright light); lack of discriminative stimuli for sleep; electronic device use; exposure to bright nightlight	Lack of physiological sleep pressure; access to food and drink; parent responses to CC's; intrusive internal stimuli; hyperarousal	Lack of physiological sleep pressure; exposure to bright light & stimulating content on electronic devices; access to food and drink; parent responses to CC's; intrusive internal stimuli; hyperarousal
Hypothesised function	Tangible Escape	Tangible Escape	Tangible	Tangible	Tangible Social attention Escape (from intrusive internal stimuli)	Tangible Social attention Escape (from intrusive internal stimuli)
Parent treatment components	Sleep hygiene; bedtime fading; unmodified extinction (removal of devices at night & scheduled device use); positive reinforcement for successive approximations towards goal	Sleep hygiene; bedtime fading; unmodified extinction (removal of devices at night & scheduled device use); positive reinforcement for successive approximations towards goal	Sleep hygiene; graduated extinction (removal of devices at night); bedtime fading; consistent sleep cues; positive reinforcement	Sleep hygiene; graduated extinction (removal of devices at night); bedtime fading; consistent sleep cues; positive reinforcement	Unmodified extinction (minimal engagement post-bedtime); bedtime fading (set sleep & wake times); positive reinforcement	Restricted access to devices after dinner; unmodified extinction (minimal engagement post-bedtime); bedtime fading (set sleep & wake times); positive reinforcement

Young person treatment components	Psychoeducation; social story; relaxation techniques; comfortable sleep setting	Psychoeducation; social story; relaxation techniques; comfortable sleep setting; Gro-Clock	Psychoeducation; social story; Finished Box; sleep item; replacement of nightlight; relaxation techniques	Psychoeducation; social story; Finished Box; sleep item; replacement of nightlight; relaxation techniques	Psychoeducation; relaxing bedtime routine; sleep checklist; relaxation techniques; imagery	Psychoeducation; relaxing bedtime routine; sleep checklist; relaxation techniques; imagery

Note. CCs = curtain calls.

During the intervention phase, the researcher communicated daily with parents. Daily (Niko) and weekly (Peter and Eric) contact was also maintained with participants. During regularly scheduled contact, the researcher provided participants with feedback regarding treatment implementation, treatment fidelity was monitored, and praise and encouragement was given. The intervention continued until the participant's sleep disturbance had been significantly reduced or eliminated, and this pattern had been evident consistently across a 10- to 14-day period. The intervention phase lasted for 48, 94, and 84 nights respectively for Niko, Peter, and Eric. FBA results and individualised treatments are detailed below.

The FBA revealed numerous antecedent and consequence variables appeared to be interfering with participants' sleep. These included lack of physiological sleep pressure due to inconsistent sleep/wake times and daytime sleep; stimulating activities pre-bedtime; presence of intrusive internal stimuli (e.g., reports of distressing cognitions); inappropriate sleep dependencies (e.g., electronic devices); lack of discriminative stimuli for bedtime (e.g., inconsistent bedtime routines); sleep environment discomfort; and exposure to light. Reinforcement contingencies for sleep-interfering behaviour came in the form of parental attention; access to electronic devices and other preferred items (e.g., food and drink); and purported escape from intrusive internal stimuli (reduced distress).

Intervention components implemented with all three participants included discussions about key sleep-facilitative behaviours (e.g., closing eyes), the impact of sleep disturbance on areas of importance to them (e.g., ability to play video games well), and connection between their behaviour and sleep disturbance. All three also received instruction in relaxation training (e.g., PMR, deep breathing) which has been suggested to reduce physiological arousal and support independent sleep onset (Stewart and Gordon, 2014). All parents received psychoeducation regarding sleep hygiene (Jan et al., 2008) and the relationship between existing operant and respondent conditioning processes and their child's sleep disturbance, as well as implemented extinction procedures (Kuhn and Weidinger, 2000), and positive reinforcement strategies.

Varying topographies and functions of sleep problems necessitated the use of additional individualised intervention components. Individualised components implemented included (a) social stories to depict new targeted sleep routines, sleep-conducive behaviour, and reinforcement contingencies (see Appendix O; Gray and Garand, 1993); (b) sleep checklists (a visual schedule of the bedtime routine; see Appendix P); (c) appropriate sleep dependencies

(provision of sleep-conducive stimuli that were accessible throughout the night e.g., a soft toy; Jin et al., 2013); (d) imagery to redirect sleep-interfering cognitions; and (e) Gro-Clocks (to provide a discriminative stimulus for sleep/wake times; see Figure 4.1). Verbal instruction, social stories, modelling, visual aids (e.g., picture cues), parent presence, and participant interests (see Appendix Q) were integrated to facilitate participants' comprehension and engagement with therapeutic resources. Individualised parent-implemented interventions included (a) bedtime fading (Piazza & Fisher, 1991a; 1991b) to increase physiological sleep pressure and create a motivating operation for sleep; (b) clear discriminative stimuli for bed preparation and sleep onset (e.g., consistent statements about bedtime and sleep); (c) scheduled access to putative reinforcers (Jin et al., 2013); (d) graduated extinction (Vriend et al., 2011); and (e) positive reinforcement for successive approximation towards desired sleep behaviour.

Figure 4.1. Picture of a Gro-clock (sourced from https://www.gro-store.com/groclock.html)

Niko. Niko's primary sleep problems included frequent and prolonged NWs as well as EWs. FBA results suggested Niko's sleep disturbance was maintained by access to tangible reinforcement (electronic device use) and escape from the perceived discomfort and cold of his bed to a heated lounge. Lack of physiological sleep pressure, due to inconsistent sleep/wake times and daytime sleep was also suggested to be contributing to Niko's sleep difficulties.

The following treatment components were implemented simultaneously: sleep hygiene; psychoeducation; a social story; a comfortable sleep environment; bedtime fading; relaxation techniques; unmodified extinction (restricted access to electronic devices and lounge heater); Gro-Clock; and positive reinforcement for successive approximation towards desired sleep behaviour (e.g., remaining in bed until successively later from 5:00am). The preceding techniques functioned to increase sleep pressure, reduce reinforcement for sleep-interfering behaviour, and promote engagement in sleep-conducive behaviour. Niko showed a reluctance to stop using electronic devices during the night, despite psychoeducation. As access to electronic devices was hypothesised to be the primary reinforcer of sleep-interfering behaviour, restricted access was imperative. Niko was taught relaxation techniques, as a replacement behaviour for leaving his bedroom when he woke, to facilitate reinitiation of sleep. Niko agreed a Gro-Clock was necessary to signal appropriate sleep/wake times as he was unable to read the time, enabling him to earn rewards. Immediate reinforcement for improvements in sleep outcomes was provided to strengthen Niko's participation in therapy, as his parent indicated he would become unmotivated and noncompliant rapidly without immediate reinforcement for progress. Reinforcement options were collaboratively agreed on by Niko and his parent.

Peter. Peter's primary sleep problems included delayed SOL and EWs thought to be reinforced by access to preferred items (e.g., electronic devices). Additional precipitating and maintaining factors included lack of physiological sleep pressure, inappropriate sleep dependencies (e.g., electronic device use), and lack of discriminative stimuli for bedtime. Intervention Phase 1 consisted of teaching Peter relaxation strategies and providing a social story. These were implemented first to provide a rationale for and prepare Peter for later changes, provide him with skills to manage anxiety, and facilitate sleep onset. These components also reduced parent anxiety regarding Peter's capacity to cope with change and thereby enhanced treatment fidelity.

Phase 2 of intervention included consistent implementation of appropriate sleep dependencies (e.g., soft toy); discriminative stimuli for sleep (e.g., switching off the bedroom light at bedtime); and graduated extinction of mobile phone use, achieved by scheduled access to all of his devices until 15 min prior to bedtime at which point he was asked to place his mobile phone in a visually enticing 'Finished Box' (see Figure 4.2), and was reinforced for compliance (immediately provided an edible treat, e.g., chocolate coin). Peter chose a sleep item (e.g., soft toy) to take to bed with him, as an appropriate sleep dependency, and his parents issued a consistent sleep statement and turned off his bedroom light. Peter's bright nightlight

was replaced with a dim one to facilitate melatonin secretion and reduce the visibility of preferred items, while continuing to provide a source of comfort. Bedtime fading was an additional component implemented to ensure sufficient physiological sleep pressure and motivating operations for sleep.

Figure 4.2. Photograph of Peter's 'Finished Box'

Intervention Phases 3 and 4 included the above components plus the graduated extinction of Peter's iPad and then laptop respectively. Reinforcement for compliance was faded as Peter learnt to independently put his devices away and go to bed on time. Phase 5 consisted of fading Peter's bedtime earlier in 15-min increments to an age appropriate time.

Eric. Eric's primary sleep problems included frequent CCs and a delayed SOL. His FBA revealed his sleep disturbance was maintained by both positive and negative reinforcement contingencies, including access to tangibles (electronic devices and food), social attention, and by sleep-interfering cognitions (e.g., "What if something happens to Mum when I'm asleep?"). Antecedent variables implicated in Eric's sleep disturbance included lack of physiological sleep pressure, hyperarousal, and exposure to bright light and stimulating electronic device content prior to bed. Exposure to bright light from electronic devices immediately before bedtime may have interfered with Eric's natural melatonin secretion. Furthermore, exposure to device content was suggested to interfere with Eric's ability to reach a relaxed state.

Intervention included simultaneous implementation of psychoeducation (instruction regarding the importance of sleep, and the impact of sleep-interfering and sleep-conducive

behaviour); a sleep checklist; restricted access to electronic devices prior to bedtime; unmodified extinction (minimal parent response to CCs); bedtime fading; and relaxation and imagery (taught to picture a pleasant/peaceful scene during sleep onset). Following consultation with the researcher, Eric agreed to stop using electronic devices after dinner and further enforcement of restrictions (i.e., an extinction procedure) was not necessary. Eric's sleep checklist supported his tendency for rule-following and bedtime routine compliance. Collaboration regarding checklist items provided Eric some control over his bedtime routine and was intended to increase his motivation to adhere to intervention. Giving him a relaxing bedtime routine, relaxation instruction, and delaying bedtime functioned to reduce the hypothesised association between bed and hyperarousal. Eric's mother was also instructed to minimise interactions post-bedtime and Eric was taught relaxation skills to facilitate independent management of sleep-interfering cognitions and reduce reinforcement for sleep-interfering behaviour.

Short- and long-term follow-up. Short- and long-term follow-up data were collected for 1 week using sleep diaries at 3 to 5 weeks and 12 to 13 weeks post-treatment. An 18- to 24-month follow-up phone interview was conducted with parents at which time they were also asked to begin a 7-day sleep diary. However, only Eric's family completed the extended follow-up diary.

Measures

Clinical interviews. Separate clinical interviews were conducted with each family and participant prior to commencing baseline. Information on past and present sleep disturbance, participant developmental history, family context, and possible environmental (e.g., bedtime routine) and mental health factors (e.g., anxiety) that could be interfering with sleep were discussed. Parental attendance at the young person's interview, incorporation of special interests, visual stimuli (e.g., sleep cartoons), minimization of direct contact (e.g., sitting side by side, use of Skype), and an open, closed, and multiple-choice question format was utilized to facilitate participant engagement.

Parent-reported sleep diaries. Parents recorded data in daily sleep diaries during each phase of the study. Diaries were used to record (a) frequency and duration of daytime sleep; (b) duration of SOL; (c) frequency of CCs; (d) frequency and duration of NWs; and (e) time of morning waking. The latter was used to calculate discrepancy between actual and goal wake

time and identify incidents of EW. Participants' sleep setting, behaviour during CCs and NWs, as well as parents' responses to this behaviour were also noted. Sleep diaries were returned to the research team on a weekly basis.

VSG. Swann Advanced-Series DVR4-1200, nighttime, infrared video cameras were used to record participant's sleep and to permit the coding of interobserver agreement (IOA) data. Information obtained from video included (a) topographies of awake behaviour post-bedtime (e.g., vocalisations, stereotypy, play); (b) topographies of sleep behaviour (e.g., sleep position, eye movement, limb movement); (c) duration of SOL; (d) frequency of CCs; (e) frequency and duration of NWs; and (f) time of morning waking. The following operational definitions were used to code video (a) asleep, lying down with minimal non-discrete movement, and no indication of wakefulness; and (b) awake, the presence of any sleep-interfering behaviour, eyes open, or frequent physical movement (Jin et al., 2013). Recording began when the participant went to bed and ended when they awoke to begin the day. Video footage was downloaded to an external hard drive and distributed regularly to the research team to enable objective monitoring of participant progress.

CSHQ. The CSHQ was completed during assessment and post-treatment to evaluate change in parent-reported sleep disturbance. The CSHQ was completed by Niko and Eric's parents. Peter's parents did not complete the CSHQ as Peter was not within the measure's validated age range. The CSHQ is a parent-report questionnaire consisting of 45 items relating to children's sleep patterns, scores > 41 are indicative of clinically significant sleep disturbance (Owens, Spirito, McGuinn, 2000).

GARS-3. The GARS-3 was used to corroborate ASD diagnoses and indicate symptom severity. The GARS-3 is a 56-item informant rating scale of autism symptomatology (Gilliam, 2013). It is designed to assess the likelihood a person has ASD and the severity of their behaviour in accordance with the DSM-5. Items are summed to provide a total ASD Index score. Higher scores indicate a high likelihood and increased severity of ASD. Probability estimates are classified as Unlikely, Probable, or Very Likely.

Treatment acceptability. The Treatment Acceptability Rating Form-Revised (TARF-R; Reimers, Wacker, Cooper, & DeRaad, 1992) was administered post-treatment to assess parents' perceptions of overall treatment acceptability. The TARF-R consists of 17 items which examine ratings of treatment acceptability and three items assessing problem severity and participants'

understanding of the intervention approach. Ratings on six subscales (Effectiveness; Reasonableness; Willingness; Cost; Negative side-effects; Disruption/time) are summed to provide a total treatment acceptability score. In addition, parents and their child were interviewed separately to assess the social validity of treatment and to provide qualitative information regarding treatment effects. Information pertaining to sleep (e.g., fatigue, sleep quality), secondary outcomes (e.g., mood), preferred and non-preferred assessment and treatment components, knowledge regarding healthy sleep habits, and suggestions for improvement were gathered. The format of the participant interview included open questions, closed questions with multiple choice options, and visual aids (e.g., photos of treatment components) to facilitate communication.

IOA (See Table 4.3). Video footage was coded by a researcher blind to parent sleep diary recordings. Agreement between parent report and direct observation data extracted from video was then calculated. Sleep phenomena which parents could not be expected to detect (e.g., covert awakenings in which the young person remained quiet in their bed) were omitted from IOA calculations. Measures of duration (e.g., SOL), sleep, and wake times were considered in agreement if parent and video were ± 15 min. IOA for CC frequency was calculated on occasions whereby bids for attention were detectable by video (e.g., clear calling out, or participant returned to bed by a parent). Percent agreement for each behaviour was calculated using the equation [Agreement/ (Agreement + Disagreement)] × 100. IOA data were collected for 22%, 46%, and 5% of nights across baseline and treatment phases for Niko, Peter, and Eric respectively. Incomplete or lack of sleep diary entries on nights when video data was available, inhibited calculation of IOA on more nights. Limited IOA data were collected for Eric as he withdrew consent for video recording on night 107 due to feeling it was an invasion of his privacy. Niko's mean IOA was 86% for duration of NWs and 100% for duration of EWs. Peter's mean IOA was 93% (range, 85–97%) for SOL and 98% (range, 86–100%) for duration of EWs. Eric's mean IOA was 95% (range, 91–100%) for CCs and 100% for SOL.

Table 4.3. *Interobserver Agreement (IOA) Between Sleep Diaries and Videosomnography Across Target Behaviours*

Target Behaviour	Niko		Peter		Eric	
	Baseline	Intervention	Baseline	Intervention	Baseline	Intervention
CCs	_	_	_	_	91%	100%
SOL	_	_	85%	97%	100%	100%
Duration NWs	_	86%	100%	100%	_	_
Duration EWs	_	100%	86%	100%	_	_

Note. Lack of baseline sleep diary data for Niko inhibited calculation of IOA in this phase.

Treatment fidelity (See Table 4.4). A checklist based on a task analysis of each measurable parent-implemented treatment component (e.g., delivery of reinforcement) was created for all families. Parent treatment fidelity was assessed by comparing data gathered from the researcher's daily contact notes, video footage, and sleep diaries with the protocol outlined in the treatment checklist. Parent treatment fidelity was calculated for 90% or more of intervention nights across all participants, using the formula (Completed tasks/Total tasks) × 100%. An aggregate score was then calculated for each participant. Treatment fidelity scores were 93%, 98%, and 71% for Niko, Peter, and Eric's parents respectively ($M = 87\%$). Niko, Peter, and Eric's parents followed every component of the treatment plan (i.e., reached 100% treatment fidelity) on 72%, 84%, and 26% of nights respectively.

Treatment fidelity for each adolescent participant was assessed by examining participant report and video for evidence of treatment adherence. The imperceptible nature of some of the components (e.g., imagery) and unreliable reporting inhibited direct measurement of treatment fidelity; however, there was indirect evidence of intervention compliance by participants. Niko demonstrated mastery of relaxation skills during intervention sessions and was able to identify when to utilise such strategies. However, he was rarely observed to apply these within the sleep context. There was little video evidence of Niko or Peter completing relaxation exercises. After Niko's access to electronic devices was restricted via password protection, he complied with the treatment plan and remained in bed, stating he closed his eyes to reinitiate sleep upon waking as opposed to utilising relaxation strategies. Initial video footage of Eric revealed he completed

PMR frequently prior to sleep onset. Eric reported using his sleep checklist and deep breathing strategies every night, although he noted that he did not always stick consistently to his bedtime routine during school holidays.

Table 4.4. *Parent Treatment Fidelity*

	Niko	Peter	Eric
Treatment fidelity	92%	98%	70%

Data Analyses

Visual analyses. Visual analysis was used to assess the effectiveness of FBA-informed interventions and additional therapies. Level, variability, and trend in the data were evaluated across study phases (Kazdin, 2001).

Effect size estimate. Visual analysis was supplemented by calculating PBM (calculated thus as behaviour decrease was therapeutic) to estimate effect sizes: < 70% represents ineffective treatment; 70 to 90% moderate effectiveness; and > 90% high effectiveness (Ma, 2009). The RCI was used to ascertain whether differences in pre- and post-CSHQ scores reflected true significant change as opposed to measurement error (Jacobson and Truax, 1991), clinical change occurred when pre-CSHQ scores reduced from the clinical range to the normal range.

Results

Treatment results for each participant are presented individually in a case study format. Sleep diary data for each participant are presented in Figures 4.3, 4.4, and 4.5.

Data Quality

Video footage was recorded on 20–88% of nights across all participants throughout baseline and treatment phases. Parents recorded sleep diary data for all dependent variables across baseline and treatment on 37–46% of nights (i.e., although parents filled out sleep diaries regularly, they did not collect information on each variable each night).

Niko's baseline sleep diary data is scarce as his parent had difficulty recording diary data during that phase, this prevented calculation of IOA in baseline. There are no sleep diary data for Peter from nights 80 to 102; Peter's parents recorded video only on these nights. Eric's SOL

sleep diary data is scarce from night 36 to 85 as school holidays resulted in variability in sleep settings (e.g., tent, friend's house).

Niko

Niko displayed high variability in the duration of NWs (0 to 120 min) and EWs (0 to 180 min) in baseline (see Figure 4.3). There was an immediate reduction in the level and variability of both sleep variables, with PBM scores (NW = 100%, EW = 95%) demonstrating a large treatment effect. NWs and EWs were eliminated by the end of treatment and these effects were maintained at both short- and long-term follow-up.

At the 24-month follow-up, Niko and his parent reported that the elimination of NWs and EWs had been maintained. Access to devices was still restricted during the night, and Niko used a watch as opposed to a Gro-Clock to signal an appropriate rise time. Additional treatment techniques were no longer required.

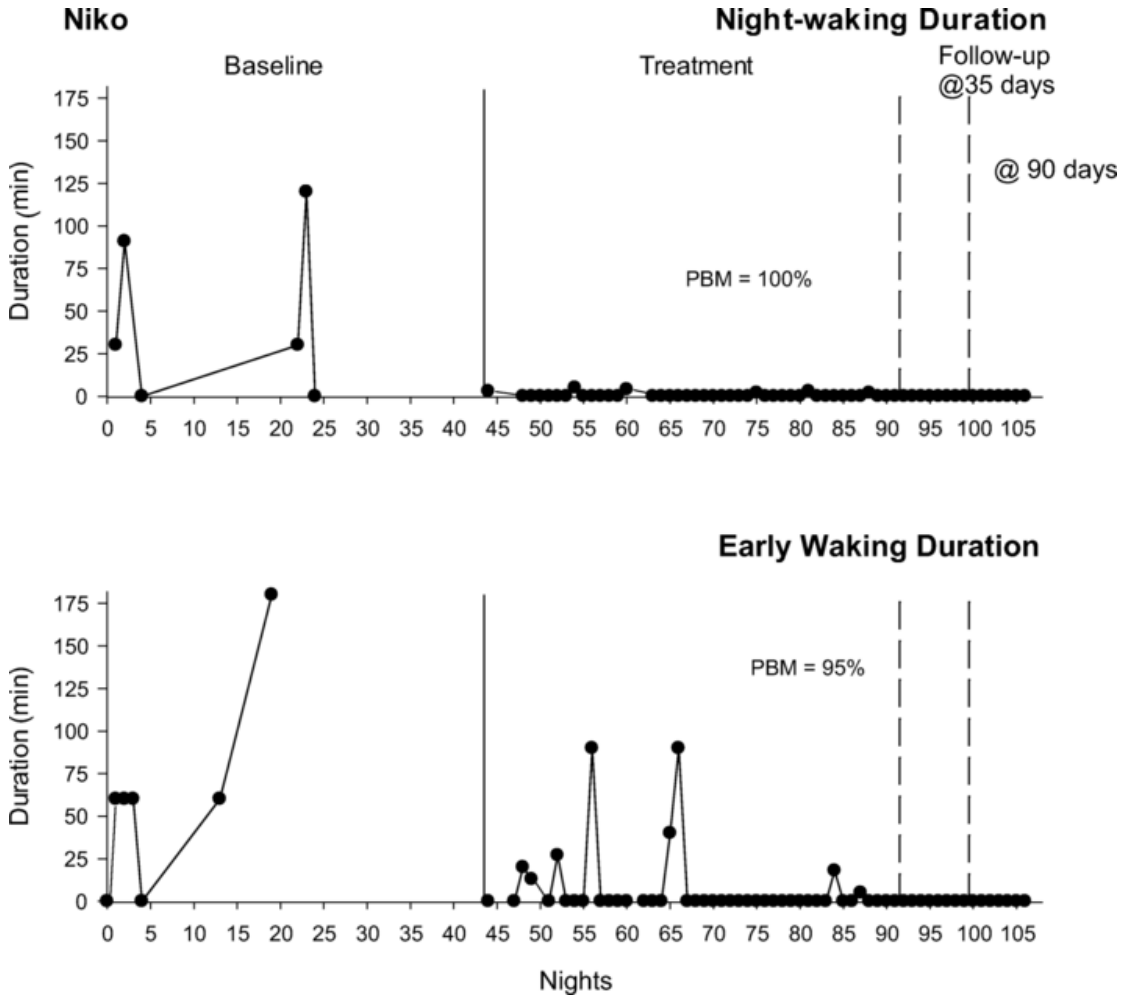

Figure 4.3. Sleep outcomes for Niko: Duration of night wakings and early wakings across baseline, intervention, and follow-up phases. PBM = Percentage below the median.

Peter

Peter's SOL was highly variable during baseline (range, 5 to 195 min; see Figure 4.4). There was an immediate reduction in the level of SOL upon implementation of relaxation strategies. The level and variability reduced further from intervention Phases 2 to 4. A large treatment effect was observed within intervention Phase 4 (PBM = 100%). SOL treatment effects were maintained at follow-up. The duration of Peter's EWs was highly variable during baseline (range, 0 to 180 min). There was an immediate reduction in the level and variability of the duration of EWs at treatment onset and they were eliminated during treatment, with this result maintained at short- and long-term follow-up.

At the 18-month follow-up, parents indicated that Peter was not experiencing sleep disturbance, SOL was 15 min, and he was not experiencing NWs or EWs. Peter's family still used the Finished Box and restricted his sleep during the day. The social story, sleep item, and reward system were no longer required.

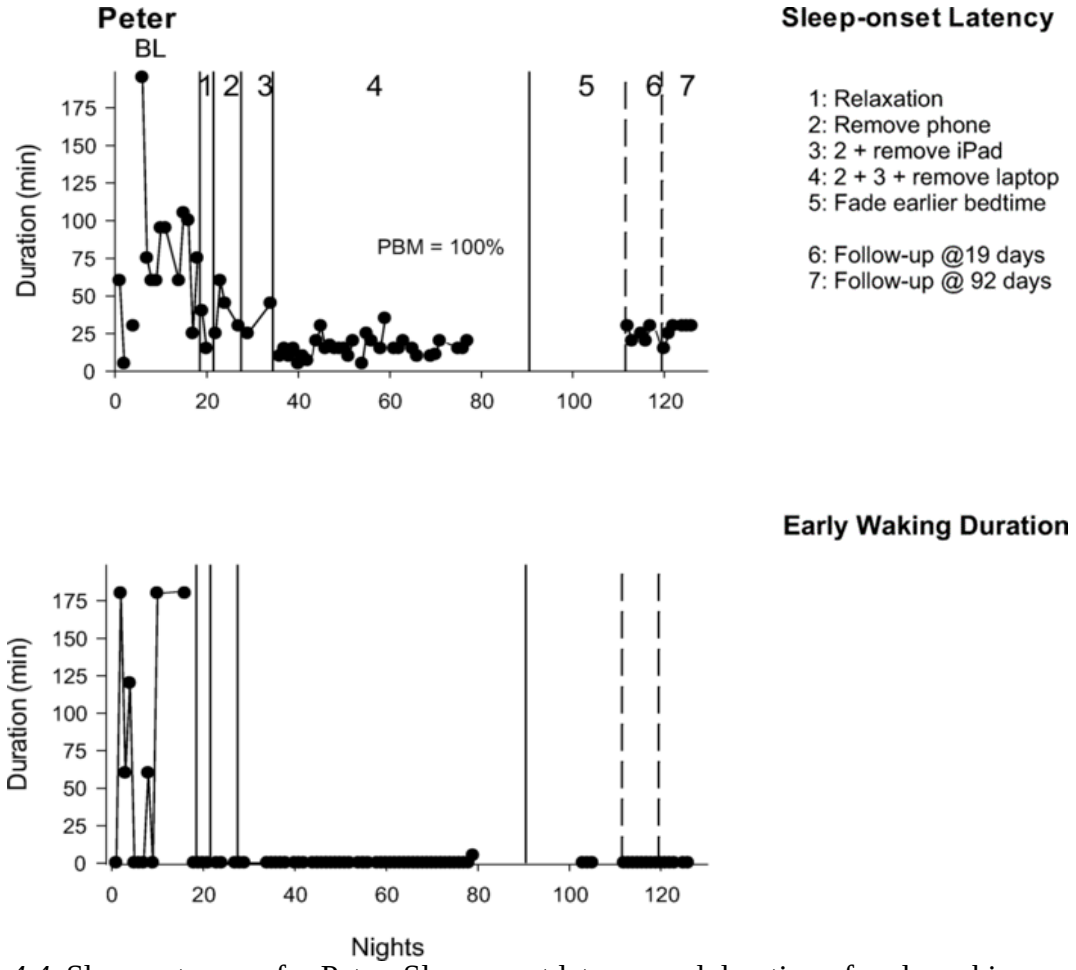

Figure 4.4. Sleep outcomes for Peter: Sleep onset latency and duration of early wakings across baseline, intervention, and follow-up phases. BL = baseline; PBM = percentage below the median.

Eric

In baseline, Eric displayed a high frequency of CCs and significant variability (range, 0 to 6; see Figure 4.5). There was an immediate reduction in the level and variability of CCs with intervention. Treatment had a moderate effect on CCs (PBM = 88.5%). The high frequency of Eric's CCs on nights 107 and 114 occurred during his transition to a new school. SOL was prolonged and highly variable during baseline (range, 15 to 450 min) but reduced significantly during intervention (PBM = 100%), and this was maintained at short- and long-term follow-up.

At 18-month follow-up, Eric's parent reported Eric was not experiencing sleep problems and no longer engaged in bedtime resistance. Eric's mother said his SOL was 10 to 15 min in duration. The family reported continued use of delayed bedtime, restricted access to devices, and use of deep breathing.

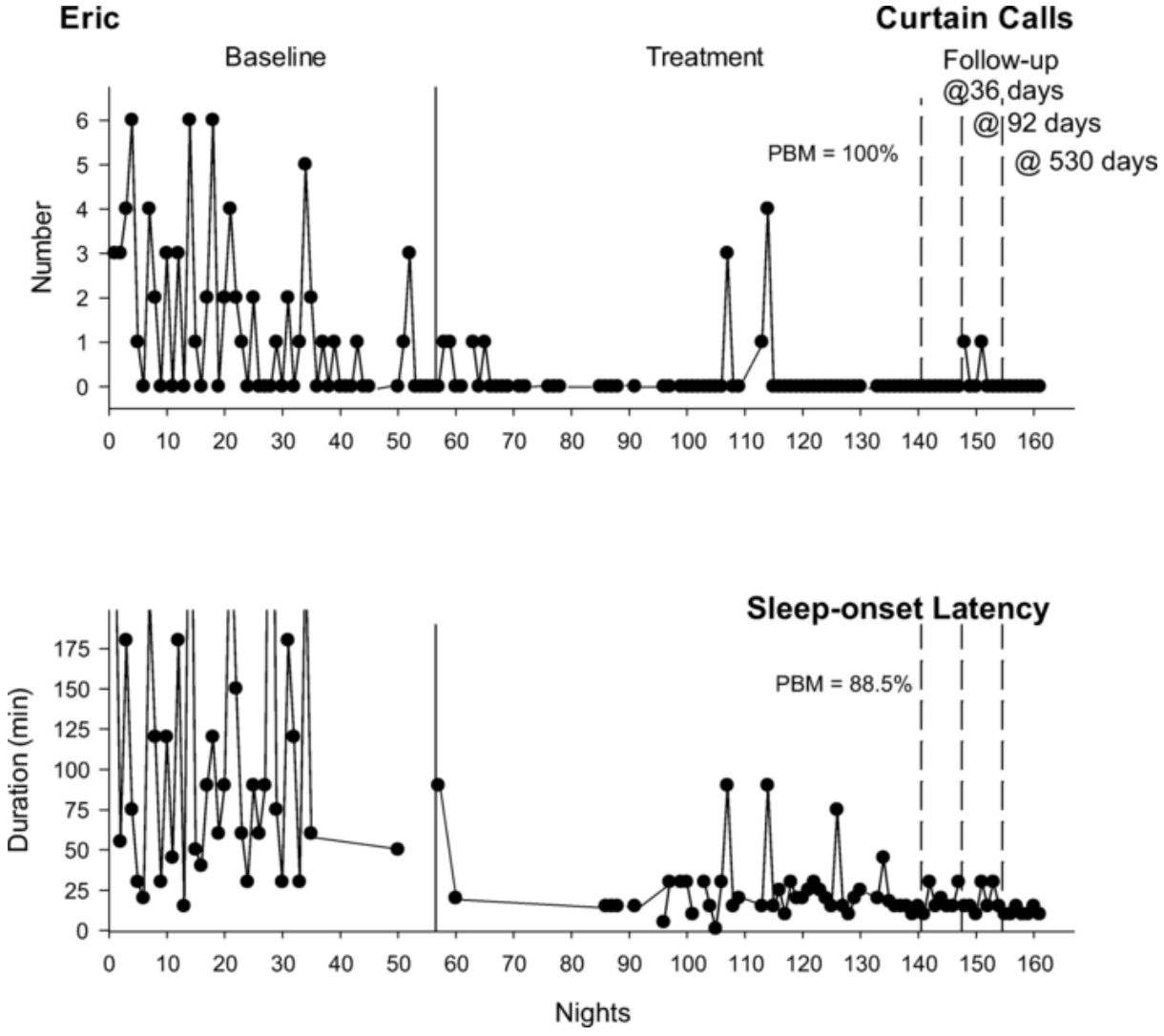

Figure 4.5. Sleep outcomes for Eric: Curtain calls and sleep onset latency across baseline, intervention, and follow-up phases. PBM = percentage below the median.

CSHQ Scores

Both Niko and Eric experienced a reliable change in their CSHQ scores (see Table 4.5). For Eric, this change was clinically significant, with his post-treatment score falling below the clinical cut-off for sleep disturbance.

Table 4.5. *Pre- and Post-treatment Children's Sleep Habits Questionnaire (CSHQ) Scores*

	Niko	Eric
Pre-treatment CSHQ	52	58
Post-treatment CSHQ	44*	38**

Note. * = significant change. ** = clinical change.

Social Validity

Participant report suggests FBA-informed sleep interventions and young person-implemented components were acceptable. Planned use of tangible and social rewards was well regarded. Participants varied in their rating of relaxation. Eric reported finding deep breathing helpful. He attributed his success to this and noted that engaging in deep breathing at bedtime helped prevent his mind from "buzzing" as he focused on counting his inhalations and exhalations. Niko and Eric reported PMR was ineffective and Peter was ambivalent. Peter was the only participant to indicate he did not like the elimination of electronic device use. Some participants commented on the intrusive nature of the video camera and the discomfort and time-consuming nature of having to talk to a researcher daily. Niko and Eric indicated they had experienced improvements in their sleep, including reduced SOL and daytime fatigue, as well as ability to reinitiate sleep upon waking.

During post-treatment interviews, all parents reported that the intervention had successfully reduced their child's sleep disturbance and perceived daytime fatigue. Importantly, parents felt their child had developed the skills to manage their sleep independently by implementing "the tools in his strategy bag" (e.g., deep breathing) and adhering to sleep-facilitative behaviour (e.g., remaining in bed). Peter's parents reported that, while visual aids were important for his comprehension, they also facilitated parental structure and routine. Eric's treatment, which consisted predominantly of intervention components delivered directly to him, was described by his parent as "non-invasive."

TARF-R

TARF-R scores have a possible range of 17 to 119; higher scores indicate higher acceptability. Parent ratings ranged from 94 to 113 (see Table 4.6). Each parent's rating yielded the maximum score on the Effectiveness subscale. Overall, parents rated the interventions to be highly acceptable, effective, and easy to understand, taking little time to implement, at no financial cost.

Table 4.6. *Treatment Acceptability Rating Form-Revised Scores*

	Niko		Peter		Eric	Maximum Score
Scale	Father	Mother	Father	Mother		
Effectiveness	21	21	21	21		21
Reasonableness	21	19	18	20		21
Willingness	21	18	16	17		21
Cost	14	10	14	14		14
Negative side-effects	21	15	15	15		21
Disruption/time	15	18	16	18		21
Problem severity*	10	11	11	2		14
Understanding of treatment*	7	7	7	7		7
Total acceptability	113	98	94	105		119

Note. * Not included in total acceptability score

Discussion

The purpose of this study was to evaluate the feasibility and effectiveness of individualised behavioural sleep interventions involving input from young people with ASD alongside parent-implemented treatment, with a focus on maintenance and social validity. Further, the validity of parent-report sleep diaries was also assessed. Overall, results of this pilot study suggest comprehensive, individualised interventions including both young person- and parent-implemented treatment components can reduce sleep disturbance. Parents reported that improvements in sleep were maintained at 18- and 24-months post-intervention. However, caution is warranted in interpreting these maintenance data because of reliance on parent report and lack of direct observation. Overall, the treatment components were rated as acceptable (socially valid) by young people with ASD and their parents. Specifically, all participants reported that they enjoyed the reinforcement systems and two reported that the young person-implemented treatment components (e.g., deep breathing) were beneficial.

Treatment effectiveness and the feasibility of young person-implemented intervention components appeared to have been mediated in part, by each participant's functioning and their use of individualised treatment strategies (e.g., relaxation). Sleep problems resolved to the greatest extent for Eric who had the highest communicative abilities and participant treatment fidelity. By contrast, Niko and Peter required additional reinforcement contingencies to increase sleep-conducive behaviour. Individuals with more severe ID may struggle to refrain from engaging in sleep-interfering behaviours that offer immediate reinforcement and may experience difficulty generating appropriate alternative responses (Ho et al., 2015). Niko attempted to gain access to electronic devices as intervention began, only engaging in sleep-conducive behaviour after access to these had been restricted. Although Eric evidenced mastery of relaxation training skills, Peter required significant support to learn relaxation skills (e.g., social story, visual aids, modelling). After four sessions, his relaxation technique still did not appear correct and he was not able to identify when to utilise the strategies. Echolalia and compromised memory and sequencing abilities appeared to limit Peter's ability to engage in conversation. Niko and Peter also each had difficulty attending to therapeutic tasks. Adolescents with ASD are less likely to initiate social interaction (Chevallier et al., 2012). Limited social skills may have contributed to the inhibited engagement with Peter and Niko.

Parents within the current study consistently reported that participants had become responsible for their own sleep and had developed skills to engage in appropriate sleep behaviour independently, likely facilitating the maintenance of treatment effects over an extended time period. Psychoeducation may have been sufficient for Eric to reduce use of electronic devices without resistance, particularly as this was not the primary maintaining reinforcer for his sleep-interfering behaviour. Furthermore, an increase in Peter's sleep-conducive behaviour (e.g., eyes closed, lying still) was observed following explicit, concrete instructions within a social story. Interestingly, he also began reprimanding other family members for sleeping during the day. Parents play an important role enforcing limits to facilitate sleep-conducive behaviour, nevertheless delivering treatment directly to young people provides them with the skills and knowledge to sustain healthy sleep practices. Treatment maintenance is critical to help prepare young people with ASD for adulthood and ensure they can function most effectively within living, education, and vocational settings without being compromised by the effects of sleep disturbance.

Parent treatment fidelity was an issue for one of the three families. Eric's parent completed every component of the treatment plan on only 26% of nights. It was particularly difficult for this family to refrain from providing social attention post-bedtime and to maintain consistent sleep and wake times respectively. In this case, low parent treatment fidelity did not appear to affect Eric's sleep outcomes, perhaps because his own treatment fidelity was high. Treatment fidelity was not a challenge for Niko and Peter's parents. The complexity of everyday life for parents of children with neurodevelopmental disorders may impact their ability to consistently focus their attention and energy on improving their child's sleep and prioritise this one aspect of their lives (Beresford et al., 2016).

VSG has rarely been used within the ASD and sleep literature; however, this objective measure can be collected within participants' homes and enables detection of salient information (e.g., topographies of sleep and awake behaviour) unobtainable through actigraph or PSG recordings (Moore et al., 2017). High IOA between parent-reported sleep diaries and VSG was found, replicating results of previous sleep research with young people with ASD (Jin et al., 2013). However, our VSG data revealed parents were often unable to detect covert sleep-interfering behaviour (e.g., early morning device use). As a result, they struggled to identify the length of sleep onset or wakings. Parent-report sleep measures may be more reliable within pre-school-aged populations, as sleep-interfering behaviour tends to

involve overt signalling to the parent or significant disruption to the household. Understandably, as Eric's case suggests, increasing desire for privacy may prevent adolescent participants consenting to the use of VSG. Further, increasing sexual desire during adolescence, paired with the lack of social understanding apparent in many individuals with ASD, may put this population at risk of being inadvertently recorded while engaging in sexual activities. Privacy issues necessitate careful consideration of VSG with any population, but particularly perhaps with adolescents.

Limitations and Future Directions

The current study illustrates treatment components directed towards young people are feasible and may be beneficial additions to traditional parent-implemented sleep interventions when working with verbal older children and adolescents with ASD. Future research aimed at determining the extent to which intellectual functioning and motivation may influence the effectiveness of sleep interventions implemented with young people appears warranted. Of consideration within the current study is the small number of participants with heterogeneous presentations of sleep problems and ASD characteristics, inhibiting generalisability to other young people with ASD. Second, recording covert sleep-interfering behaviour via parent report may not be sufficiently accurate, suggesting a need for research aimed at evaluating the veracity of dependent variables used in sleep-intervention research. Third, the single-case design used did not isolate the effects of any specific intervention component and it is not possible to determine whether any given component was necessary or sufficient. However, the results of this pilot study suggest future research could include a component analysis and experimental design capable of demonstrating experimental control.

Chapter 5: Study 2

Behavioural Treatment of Sleep Disturbance in Children and Adolescents with Autism[2]

Children and adolescents on the autism spectrum have a higher incidence of sleep disturbance than their typically developing counterparts (Deliens, Leproult, Schmitz, Destrebecqz, & Peigneux, 2015). A recent large-scale study found evidence of clinical sleep problems amongst 74% of the 3- to 18-year-olds with ASD in their sample (Goldman et al., 2012). This compares to prevalence rates of 9 to 50% in typically developing young people (Richdale & Schreck, 2009). Sleep problems are associated with numerous detrimental impacts, including deficits in cognitive and adaptive functioning, increased autism symptom severity, challenging behaviour, and poor emotional regulation (Goldman et al., 2011; Taylor et al., 2012; Rzepecka et al., 2011). It also detrimentally effects parent sleep quality, psychological wellbeing, and marital relationships (Hodge et al., 2013). Without effective intervention, sleep problems faced by people with ASD are unlikely to remit (Sivertsen et al., 2012).

The high rate of sleep problems in ASD is thought to be due to a complex interaction between physiological (e.g., dysregulated melatonin, comorbid medical conditions), psychological (e.g., anxiety), and behavioural (e.g., inappropriate or insufficient stimulus control of sleep-onset) factors. Given the complex aetiology of sleep problems in children with ASD, it is unsurprising that a range of treatment options have been empirically investigated. Those with the most empirical support include pharmacological (i.e., melatonin) and behavioural approaches. Behavioural sleep interventions are based on operant learning theory and typically involve bringing sleep under appropriate stimulus control (e.g., positive bedtime routines), and modifying contingencies of reinforcement for sleep-interfering/sleep-conducive behaviour (e.g., extinction procedures), as well as motivating operations for falling asleep (e.g., altering the sleep/wake schedule). FBA is a tool used to identify controlling variables that may evoke or maintain a behaviour by identifying the antecedent variables that occasion sleep problems and consequence variables that reinforce the sleep problem. This information is then used to ascertain the function of the sleep-interfering behaviour (i.e., why it is occurring), and formulate function-matched intervention plans (Blampied, 2013a; Kodak

[2] An article based on this study is under revision as requested by the editor of Research in Autism Spectrum Disorders: van Deurs, J. R., France, K. G., McLay, L. K., & Blampied, N. M. (2020). Behavioral treatment of sleep disturbance in children and adolescents with autism: A nonconcurrent multiple baseline design study. *Research in Autism Spectrum Disorders*.

& Piazza, 2008). An emerging body of research suggests that FBA-informed, parent-implemented interventions are effective at alleviating sleep problems experienced by children with ASD (Jin et al., 2013; McLay, France, Knight et al., 2019; McLay, France Blampied et al., 2019).

Within extant research, behavioural sleep interventions for young people with ASD have primarily been implemented by parents. This is in contrast to sleep interventions for typically developing children and adolescents, whereby young people are often actively included in the treatment process (e.g., by psychoeducation with dolls, coping self-talk, and/or bibliotherapy; Fehr et al., 2016; Kahn et al., 2017; Rafihi-Ferreira et al., 2018). There are many benefits to this, such as upskilling the young person to independently manage their sleep (Schlarb et al., 2011), potentially limiting the need for more restrictive or parent-implemented intervention practices (e.g., unmodified extinction). Nevertheless, young people may not consider existing sleep-interfering behaviour (e.g., device use) to be problematic, potentially compromising treatment compliance and outcomes (Cain et al., 2011). Further, many modifications to standard interventions are necessary to facilitate application to young people with ASD (e.g., adapted communication methods; short, structured sessions; incorporation of special interests). Although adapted cognitive behavioural therapies are effective for a range of psychological issues in ASD (Attwood & Scarpa, 2013; Moree & Davis, 2010; Walters et al., 2016), few studies have investigated their application to sleep disturbance (Loring et al., 2016; McCrae et al., 2019; Souders et al., 2017; van Deurs et al., 2019). As yet, there is scant research into how young people with ASD can be assisted to understand their own sleep problems and participate in the selection and implementation of any FBA-informed treatment. One recent study (van Deurs et al., 2019) provides a demonstration of how FBA-informed sleep interventions can be directly implemented with young people with ASD. While parent support for interventions was required to varying extents, this study demonstrated the feasibility of including young people with ASD in the therapeutic process, and the effectiveness, acceptability, and social validity of this approach.

While there is some evidence of the effectiveness of this approach, numerous questions remain. The efficacy of FBA-informed interventions for sleep problems in older children and adolescents with ASD has not been established, nor has the long-term effectiveness of this approach. Further, it is not known to what extent parent-implemented treatment components are necessary or whether sleep disturbance can be reduced via young

person-implemented interventions alone. Additionally, we know little about the young person's viewpoint concerning the social validity of behavioural interventions for sleep problems. The purpose of the current study was to extend the work of van Deurs et al. (2019) by evaluating (a) the effects of young-person and parent-implemented FBA-informed interventions in a larger sample of older children and adolescents with ASD; (b) the maintenance of treatment effects; and (c) the social validity of such interventions.

Method

Participants

Inclusion criteria were (a) formal diagnosis or features of ASD as verified by a psychiatrist, psychologist, or paediatrician, and supported by a high probability estimate (Very Likely) and severity level rating on the GARS-3; (b) parent-reported unwanted co-sleeping or difficulty initiating and maintaining sleep, supported by systematic in-home measurement; (c) no medical condition that directly interfered with sleep; and (d) sufficient receptive and expressive communication skills to engage in assessment and treatment processes (e.g., ability to attend to verbal language and meaningfully reply, can understand and follow simple instructions). The communication criterion was assessed through clinical interviews and observation coupled with responses to items on the Communication sub-domains of the Vineland Adaptive Behavior Scales Third Edition, (Vineland-3; Sparrow, Cicchetti, & Saulnier, 2016), e.g., "Follows directions to do something with one object" and "Says at least 50 words". Referrals to the study were initiated by parents or by ASD service providers. Eight males aged 9 to 15 years (M = 12 years) met these criteria (See Table 5.1 for a summary of participant characteristics).

Measures

GARS-3. The GARS-3 was completed by parents during assessment and used to corroborate ASD diagnoses and indicate symptom severity. The GARS-3 is a 56-item informant rating scale of autism symptomatology (Gilliam, 2013). It is designed to assess the likelihood a person has ASD and the severity of their behaviour in accordance with the DSM-5. Items are summed to provide a total ASD Index score. Higher scores indicate a high likelihood and increased severity of ASD. Probability estimates are classified as Unlikely, Probable, or Very Likely.

Clinical interviews. Semi-structured clinical interviews were conducted separately with participants and their parents during assessment. Information pertaining to sleep concerns, possible contributing factors, participant's interests, developmental history, and

Table 5.1. *Summary of Participant Characteristics at Commencement of Intervention*

Participant	Age (Y-M)	Sex	Nationality	SES	Education	Diagnosis	Medication	Receptive/ Expressive/ Written Communication Age Equivalent	Anxiety Probability
Blair	9-0	Male	NZ/ Euro	37	Home school	ASD, Chromosomal duplication 15q13.3	Atomoxetine, Risperidone	1-3 3-1 4-3	-
Seth	9-6	Male	NZ/ Euro	77	Mainstream school	ASD, ADHD, TS, APD	-	2-10 10-6 11-9	High Probability
Will	11-6	Male	NZ/ Euro	37	Mainstream school	ASD	-	4-0 4-8 10-0	Very High Probability
Finn	11-11	Male	NZ/ Euro	72	Mainstream school	Features of ASD, ADHD, Dyspraxia	Melatonin	7-3 4-2 8-4	Very High Probability

Ben	12-2	Male	South African	56	Mainstream school	ASD, GAD	Melatonin	3-5 5-10 7-6	High Probability
Scott	12-4	Male	NZ/ Euro	48	Mainstream school (teacher aide support)	ASD, ID, Dyspraxia	Melatonin, Fluoxetine	1-7 3-2 7-0	Very High Probability
John	14-2	Male	NZ/ Euro	72	Mainstream school	ASD, ADHD, TS, Dyslexia, Irlen Syndrome, APD	Melatonin, Clonidine, Atomoxetine	4-4 5-6 7-0	Very High Probability
Isaac	15-7	Male	NZ/ Māori	44	Mainstream school	ASD	-	22-0 21-0 20-0	Low Probability

Note. SES = Socioeconomic status; NZ/ Euro = New Zealand/ European; ASD = autism spectrum disorder; ADHD = attention-deficit/hyperactivity disorder; APD = auditory processing disorder; GAD = generalised anxiety disorder; ID = intellectual disability; TS = Tourette's Syndrome. SES assessed by the New Zealand Socio-economic Index 2013 (NZSEI-13; Fahy et al., 2013), based on the occupation of the principle income earner. Ten = lowest SES and Ninety = highest SES. Age equivalent communicative abilities were assessed using the Vineland Adaptive Behavior Scales Third Edition (Vineland-3; Sparrow et al., 2016). Anxiety probability was assessed using the Multidimensional Anxiety Scale for Children 2nd Edition (MASC-2; March, 2012).

family context was discussed. Parental attendance at the young person's interview, incorporation of special interests, visual stimuli (e.g., sleep cartoons), minimization of direct contact (e.g., sitting side by side, use of Skype), metaphors (e.g., magic wand to address sleep disturbance), and an open, closed, and multiple-choice question format was utilized to facilitate participant engagement. Motivational interviewing techniques were used within participant interviews to ascertain the young person's confidence and commitment to attaining sleep goals.

Behavioural Intentions Questionnaire (BIQ; Moseley & Gradisar, 2009). An adapted version of the BIQ was completed by participants during assessment and post-treatment to evaluate their motivation to engage in sleep-conducive behaviour. Using a 5-point Likert scale, participants rated their intention to engage in four or five specific target behaviours associated with their individualised treatment plan (e.g., "Go to bed at about 8:30pm every night – even on weekends"). Participants could choose from the following response options, based on the Transtheoretical Model of Behaviour Change: I don't plan to; I want to; I need to; I will; and I tried to. In addition, the following were administered during the assessment phase.

SATT. The SATT is a semi-structured interview for identifying factors underlying children's sleep disturbance. This was used to guide the clinical interview.

QABF. The QABF is a 25-item rating scale designed to assist in the identification of a target behaviour's function (e.g., social attention, escape, or tangible reinforcement). This was used to facilitate FBA.

Multidimensional Anxiety Scale for Children 2nd Edition (MASC-2; March, 2012). The MASC-2 is a 50-item questionnaire designed to assess anxiety symptoms (e.g., Social Anxiety, Separation Anxiety, Physical Symptoms) experienced by people aged 8- to 19-years. It was completed by parent's pre-treatment to evaluate the contribution of anxiety to participants' sleep disturbance. The number of elevated scores across Anxiety Scales yield an Anxiety Probability score indicating the likelihood of an anxiety disorder (Low, Borderline, High, or Very High). The MASC-2 has high reliability and validity (March, 2012).

Parent-report sleep diaries. Parent-report sleep diaries were completed daily across study phases by all except Isaac's parents. They recorded the frequency and duration of

daytime sleep; SOL; frequency of CCs; frequency and duration of NWs; and time of morning waking. Participants' sleep setting, sleep-interfering behaviour, and parents' responses to this behaviour were also recorded.

Self-report sleep diaries. Sleep diaries were completed by Isaac, who had strong reading and writing skills, confirmed by the Written subdomain of the Vineland-3 Communication scale, and who, unlike other participants, was primarily responsible for his own sleep. Isaac documented the duration of SOL; cognitions prior to sleep onset; frequency and duration of NWs; behaviour during NWs; and time of morning waking. The remaining participants used individually formatted diaries (e.g., visual stimuli only, multiple-choice) to record sleep information to supplement parent report.

VSG. VSG was used to directly observe participants' sleep across study phases. Swann Advanced-Series DVR4-1200, nighttime, infrared video cameras were placed in consenting participants' bedrooms and turned on when the participant went to bed and turned off when they awoke to begin the day. Information obtained from video replicated that obtained via sleep diaries, while also allowing for the analysis of topographies of behaviour across sleep states. The following operational definitions were used to code video: (a) asleep, lying down with minimal non-discrete movement, and no indication of wakefulness; and (b) awake, the presence of any sleep-interfering behaviour, eyes open, or frequent physical movement (Jin et al., 2013). Video was coded by a researcher who was blind to sleep diary recordings. Lack of available equipment or participant consent prohibited video collection for Seth and Will, and during follow-up for Blair, Finn, and Ben.

IOA. IOA was calculated by comparing parent-reported sleep diary data and VSG. Aspects of sleep which parents could not be expected to detect (e.g., covert wakings in which the participant remained quiet in their bed) were omitted from IOA calculations. IOA for CC frequency was calculated on occasions whereby bids for attention were detectable by video (e.g., clear calling out, or participant returned to bed by a parent). Measures of duration (e.g., SOL) and sleep/wake times were considered in agreement if sleep diaries and VSG were within ± 15 min. Percent agreement was calculated using the equation [Agreement/ (Agreement + Disagreement)] × 100. On average, IOA data was collected for 20% of nights (range, 17-22%) across target behaviours and study phases. Mean IOA was 80% (range 25-100%).

Participant and parent treatment fidelity. Fidelity was assessed by comparing measurable events recorded in daily contact notes, video footage, and sleep diaries with the prescribed treatment protocol. An aggregate treatment fidelity score was calculated using the formula (Completed tasks/ Total tasks) × 100. Participant and parent treatment fidelity was calculated on an average of 87% (range, 52 – 100%) and 70% (range, 52 – 100%) of nights respectively.

Parent treatment fidelity data was unobtainable for Will, Isaac, Finn, and Ben during follow-up due to an absence of parent input during these phases. There was insufficient information to calculate Seth's parent's treatment compliance. No participant treatment fidelity data were available for Blair and Scott, as treatment consisted primarily of parent-implemented components and young person-implemented components (e.g., controlled breathing) were not able to be reliably detected by VSG.

Overall, young person treatment fidelity was high (*M* = 84%, range, 64-93%), but tended to reduce during short- (*M* = 49%, range, 0-79%) and long-term follow-up (*M* = 57%, range, 0-86%). Similarly, parent treatment fidelity was high during intervention (*M* = 86%, range, 65-100%) and reduced during short- (*M* = 39%, range, 0-100%) and long-term follow-up (*M* = 60%, range, 0 -100%). Individual treatment fidelity scores are presented in Tables 5.2 and 5.3.

Table 5.2. *Participant Treatment Fidelity*

	Blair	Seth	Will	Finn	Scott	Ben	John	Isaac
Treatment	-	89%	85%	92%	-	80%	64%	93%
Short-term follow-up	-	0%	67%	73%	-	79%	17%	57%
Long-term follow-up	-	0%	79%	62%	-	50%	67%	86%

Note. Blair and Scott's intervention consisted primarily of parent treatment components and implementation of young person treatment components (e.g., controlled breathing) were not able to be reliably measured.

Table 5.3. *Parent Treatment Fidelity*

	Blair	Seth	Will	Finn	Scott	Ben	John	Isaac
Treatment	72%	-	-	100%	95%	98%	65%	-
Short-term follow-up	79%	-	-	-	100%	-	0%	-
Long-term follow-up	79%	-	-	-	100%	-	0%	-

Note. No parent treatment components were included in Will or Isaac's sleep intervention and had ceased for Finn and Ben during follow-up. There was insufficient information to calculate Seth's parent's treatment fidelity.

Social validity. Individual interviews were conducted with participants and their parent's post-treatment to evaluate social validity. In addition, participants and their parents completed the Young Person Treatment Evaluation (YPTE; see Appendix R) and TARF-R post-treatment respectively. The YPTE was developed for this study, based on the Child Evaluation Inventory (CEI; Kazdin, 1984). The YPTE consists of six items on a 3-point Likert scale (e.g., 1 = Not at all helpful; 2 = OK; and 3 = Very helpful) which assess effectiveness, enjoyability, fairness, time required, and overall perception of treatment. Ratings are summed to provide a total acceptability score. Scores have a possible range of 6 to 30, with higher scores indicating higher treatment acceptability. Parents completed the TARF-R to evaluate their perception of the intervention. The TARF-R is a 20-item questionnaire with six subscales (Effectiveness; Reasonableness; Willingness; Cost; Negative side-effects; Disruption/time) summed to give a total treatment acceptability score. Scores have a possible range of 17 to 119, with higher scores indicating higher acceptability.

Design

A non-concurrent multiple-baseline design across participants and target behaviour with random allocation to baseline lengths was intended to be used with all participants. Modifications to treatment necessitated by rate and level of progress were implemented with

five participants (indicated by numerical values 1, 2, 3, 4, 5, or 6 on figures 5.1 to 5.5). An AB design was used for Will, Seth, and John, an ABC design for Blair, Ben, and Isaac, an ABCDE design for Finn, and an ABCDEFG design for Scott.

Procedures

Setting. Participants lived in rural and urban areas across NZ. Eligibility was evaluated by phone prior to assessment and questionnaires were then sent to eligible participants. Pre- and post-treatment assessment interviews, treatment planning, and progress discussions were conducted in a university-based clinic, at participants homes, or via Skype. Treatment was implemented within the home by participants and their parents with remote support from the authoring intern psychologist or a registered psychologist from the wider sleep research team. Post-treatment questionnaires were sent to families at conclusion of treatment.

FBA. Clinical interviews, the SATT, QABF, sleep diaries, and analysis of video footage was used to conduct the FBA. Information gathered about the history and type of sleep problems, antecedent and consequence variables maintaining the sleep problem, and its possible function was synthesised in an FBA-informed case conceptualisation which guided treatment planning (Blampied, 2013a).

Baseline. Baseline commenced following completion of the FBA for all participants, excluding Will. Participants were randomly assigned a baseline length of 2, 3, or 4 weeks. All families were asked to maintain their existing sleep habits during this phase, although Finn and John's melatonin use was modified. Will's baseline data was collected during FBA as target behaviour improved when he was informed of his upcoming clinical interview and again following this interview. Finn and John's baseline comprised of two conditions, with (+M) and without (-M) melatonin to compare the effectiveness of melatonin versus behavioural intervention on target behaviour.

Intervention. Comprehensive, individualised, FBA-based interventions commenced upon conclusion of baseline. Details of participants' sleep disturbance, the precipitating and maintaining factors, hypothesised functions, and treatment components are summarised in Table 5.4.

Table 5.4. *Problem Behaviour, Predicted Factors Precipitating and/or Maintaining Behaviour, Hypothesised Function, and Young Person- and Parent-implemented Treatment Components for All Participants*

	Sleep disturbance	Precipitating and maintaining factors	FBA	Young person- implemented treatment components	Parent- implemented treatment components
Blair	NWs	Nightmares; cognitive and physiological hyperarousal; access to food, drink, activities; social attention	Escape Attention Tangibles	Psychoeducation; social story; relaxation; imagery; protective item (teddy); Gro-Clock; bravery certificate	Psychoeducation; unmodified extinction (minimal engagement post-bedtime); positive reinforcement
Seth	CCs Delayed SOL	Cognitive and physiological hyperarousal; access to food and activities; social attention	Escape Attention Tangibles	Psychoeducation; social story; sleep checklist; relaxation training; protective item (soft-toy duck)	Unmodified extinction (removal of books from bedroom using a "Finished Box"); positive reinforcement
Will	Unwanted co-sleeping	Social attention; inconsistent sleep cues; cognitive and physiological hyperarousal	Attention Escape	Intervention session (psychoeducation, motivational interviewing techniques, relaxation instruction)	None
Finn	CCs Delayed SOL	Inconsistent sleep cues; access to activities; social attention; cognitive and physiological hyperarousal;	Attention Tangibles Escape	Phase 1) Psychoeducation; removal of salt lamp and fairy lights; VSM; sleep checklist; relaxation training; bedtime pass; reward contract Phase 2) No radio post-bedtime Phase 3) Imagery; psychoeducation Phase 4) Encourage not to use bedtime pass	Phase 1-3) Reward contract/ positive reinforcement Phase 4) Increased reinforcement for no pass use
Ben	CCs NWs	Cognitive and physiological hyperarousal; social attention	Escape Attention	Phase 1) Psychoeducation; social story; sleep checklist; relaxation training Phase 2) Guided relaxation tape; bedtime pass	Phase 1-2) Positive reinforcement

Scott	Unwanted co-sleeping	Inconsistent sleep cues; access to devices; social attention; cognitive and physiological hyperarousal	Attention Tangibles Escape	Phase 1) Social story; relaxation training	Phase 1) Instruct to go to bedroom; positive reinforcement Phase 2-3) Fading parent presence (bedroom); positive reinforcement Phase 4) Fading parent presence (hallway); positive reinforcement Phase 5) Fading parent presence (parent bedroom, out of sight); positive reinforcement Phase 6) No parent presence; positive reinforcement
John	CCs Delayed SOL	Social attention; access to activities; intrinsic reinforcement; inconsistent sleep schedule; lack of sleep pressure; cognitive and physiological hyperarousal	Attention Tangibles Automatic Escape	Psychoeducation; bedtime fading; consistent sleep schedule; sleep checklist; self-monitoring	Quality parent-child time prior to bedtime; positive reinforcement
Isaac	Delayed SOL	Inconsistent sleep schedule; lack of sleep pressure; access to activities; cognitive and physiological hyperarousal	Tangibles	Phase 1) Psychoeducation; bedtime fading; consistent sleep schedule; stimulus control; relaxation training Phase 2) Fade bedtime earlier; address sleep inertia (e.g., splashing limbs with cold water)	None

Note. CCs = curtain calls; SOL = sleep onset latency; NWs = night waking; VSM = video-self modelling.

FBA indicated a range of factors were contributing to participant sleep disturbance, including inconsistent sleep schedules (e.g., rising late during weekends versus weekdays), lack of discriminative stimuli for sleep (e.g., inconsistent events leading up to bedtime), inconsistent availability of sleep cues (e.g., discriminative stimuli for sleep present at bedtime, such as the presence of a parent, were absent during NWs), lack of physiological sleep pressure (e.g., delayed sleep phase), and signs of cognitive and physiological hyperarousal (e.g., rumination on ideas or concerns, stimulation from engaging activities). Behaviour that interfered with sleep (e.g., leaving the bedroom post-bedtime) was thought to be maintained via the following reinforcement contingencies: access to social attention, access to tangibles (e.g., electronic devices, food), sensory stimulation (e.g., humming), and escape from distressing cognitions (e.g., dwelling on a nightmare).

Intervention was individualised for each participant and consisted of either young person-implemented treatment only (Will and Isaac) or combined young person- and parent-implemented treatment. Components were selected in accordance with FBA results, participant communicative ability and motivation, as well as family preference.

Common young person-implemented treatment components included psychoeducation, relaxation instruction, and social reinforcement. Participants were provided psychoeducation about sleep in general and the impact of their existing behaviour on their sleep problems. Based on high levels of anxiety (as indicated by MASC-2 scores), reports of fear/frustration post-bedtime, and a propensity to ruminate once in bed, participants (excluding John) were taught relaxation strategies. These included imagery, PMR, and diaphragmatic breathing. Social reinforcement (e.g., praise, certificates, joke telling, and performing magic tricks or science experiments) was also provided regularly by the researcher, contingent on engagement in sleep-conducive behaviour.

Individualised young person-implemented interventions included (a) social stories outlining participants' new bedtime routines, treatment strategies, and reinforcement systems (Burke et al., 2004); (b) VSM of the bedtime routine and sleep-conducive behaviour (Bellini & Akullian, 2007); (c) a sleep checklist (pictorial depiction of the bedtime routine) to facilitate bedtime routine compliance and consistency; (d) bedtime fading and consistent sleep/wake schedules to increase motivating operations for sleep (Delemere & Dounavi, 2018); (e) a Gro-Clock to signal appropriate sleep/wake times; (f) a bedtime pass (an individualised card enabling participants to leave their bedroom once post-bedtime, parents

responded as they would typically to subsequent bids for attention) to facilitate self-control over whether to seek parental attention and enhance independent sleep onset (Freeman, 2006); (g) no stimulating activities (e.g., listening to radio) post-bedtime to reduce hyperarousal and strengthen the association between bed and sleep (Bootzin & Stevens, 2005); (h) negotiated removal of night lights to reduce visibility of and help shift attention away from preferred items; (i) a protective item (e.g., teddy) to facilitate independent management of nighttime fears (Kushnir & Sadeh, 2012); (j) self-monitoring of sleep behaviour (e.g., CCs, SOL) with parent-delivered reinforcement for sleep-conducive behaviour (Carr, 2016); and (k) strategies to address subjective sleep inertia (e.g., splashing limbs with cold water, listening to upbeat music upon waking; Kaplan, Talavera, & Harvey, 2018). The strategies used with each participant are outlined in Table 5.4.

All participants (excluding Will and Isaac) required parent-implemented reinforcement to encourage engagement in sleep-conducive behaviour. Additional individualised parent-implemented components were necessary for Blair, Seth, Scott, and John because Seth resisted stopping engaging in sleep-interfering activities post-bedtime and due to the strong reinforcement of sleep-interfering behaviour by parent attention for Blair, Scott, and John. Individualised components included psychoeducation regarding appropriate sleep hygiene practices; unmodified extinction (e.g., restricting access to tangibles post-bedtime); systematic fading of parent presence (distancing the location between parent and child over successive nights; Howlin, 1984); and scheduled parent-child time pre-bedtime to satiate desire for parent attention post-bedtime. Any modifications to intervention conditions (see Table 5.4) occurred after at least one week had elapsed without significant change in target behaviour. These were as follows:

Finn. Access to the radio post-bedtime was eliminated on night 34 (Phase 2). However, during this phase Finn began to read post-bedtime using torchlight. Consequently, he was provided psychoeducation about the impact of this on his total sleep time and taught imagery (e.g., imagine peaceful scene) to distract himself from sleep-interfering cognitions (night 56, Phase 3). Finally, on night 68 (Phase 4) reinforcement was introduced for no bedtime pass use, in an effort to eliminate CCs.

Ben. On night 36, (Phase 2) Ben was provided with an individualised guided relaxation tape to facilitate independent use of relaxation strategies, as well as a bedtime pass to reduce CCs.

Scott. Phases 2 to 6 consisted of systematically fading parent presence from the bedroom at sleep onset and during NWs. Fading occurred on night 37 (Phase 2), night 54 (Phase 3), night 68 (Phase 4), and night 75 (Phase 5), until parent presence was eliminated on night 89 (Phase 6).

Isaac. Once SOL had stabilised on night 43 (Phase 2) Isaac's bedtime was faded earlier to increase total sleep time. He was also taught strategies to reduce sleep inertia.

Participants and their parents were contacted daily to weekly by the researcher to facilitate effective treatment delivery and monitor fidelity. Intervention continued until there was a significant reduction in sleep disturbance evidenced over a 14-day period. Mean intervention length was 61 days (range, 28-106).

Short- and long-term follow-up. Short- and long-term follow-up video and sleep diary data were collected for 1 week at *M* = 7 weeks (range, 3-9) and *M* = 15 weeks (range, 12-20) post-treatment.

Data Analyses

Visual analysis of graphed multiple-baseline data (stacked according to target behaviours) was supplemented by PBM (calculated thus as behaviour decrease was therapeutic; Ma, 2009), to evaluate treatment effect. PBM is an effect-size measure for single-case data; < 70% represents ineffective treatment; 70 to 90% moderate effectiveness; and > 90% high effectiveness (Ma, 2009). For all participants (excluding John and Finn), PBM effect sizes were calculated by comparing the baseline median to the individual data points within the final intervention phase and follow-up conditions. For John and Finn, the baseline median without melatonin (-M) was compared to the final intervention phase and follow-up conditions. The baseline median with melatonin (+M) was compared to treatment conditions to evaluate the effect size of pharmacological and/or behavioural interventions. PBM was calculated using sleep diary data, though, when diary data was missing or discrepant, video data was used (indicated in the results).

Results

Nights in which illness occurred have been omitted from Figures 5.1 to 5.5, and from the data analysis. Although parents were asked to record sleep diaries each night, occasionally diaries were completed inconsistently or included missing values. When possible, video observations were recorded on nights with missing diary data to ensure sufficient information was obtained on target variables across study phases. For John, missing SOL values within the Baseline +M phase precluded comparisons across subsequent study phases, however, effect size estimates were able to be calculated based on video observations for this variable. Treatment effects were evaluated according to video observations for Blair, as parent-reported and video-observed data were highly discrepant.

CCs

Frequency of CCs (see Figure 5.1) reduced significantly for all three affected participants by the end of treatment (Ben PBM = 93%, John PBM = 95%, Finn PBM = 100%), with improvement maintained at short-term follow-up (Ben PBM = 100%, John PBM = 100%, Finn PBM = 100%). Treatment effects were maintained at long-term follow-up for Ben (PBM = 100%) and Finn (PBM = 100%), but to a lesser extent for John (PBM = 67%). Although Finn's CC frequency increased at long-term follow-up, it remained below baseline levels.

The frequency of John's CCs during the melatonin + behavioural treatment condition was significantly lower than the melatonin alone condition (PBM [based on Baseline +M] = 100%). The frequency of Finn's CCs was significantly less during behavioural treatment alone compared to melatonin alone (PBM [based on Baseline +M] = 98%).

SOL

By the end of treatment there was a moderate to significant reduction in SOL for all four affected participants (see Figure 5.2; John PBM_{Video} = 84%, Isaac PBM = 93%, Seth PBM = 82%, Finn PBM = 92%). Treatment effects were maintained at short-term follow-up for John (PBM_{Video} = 100%), Isaac (PBM = 100%) and Finn (PBM = 100%) and long-term follow-up for Isaac (PBM = 100%) and Finn (PBM = 86%). Lack of video recordings precluded measurement of SOL at long-term follow-up for John. Treatment effects were not maintained at short- (PBM = 29%) or long- (PBM = 0%) term follow-up for Seth.

There was no significant additional effect of melatonin + behavioural treatment on John's SOL compared to melatonin alone (PBM_{Video} Baseline M+ = 56%). For Finn, there was no significant difference in the treatment effect size produced by behavioural treatment compared with melatonin alone (PBM Baseline M+ = 16%). However, Finn's SOL stability only improved during the behavioural intervention.

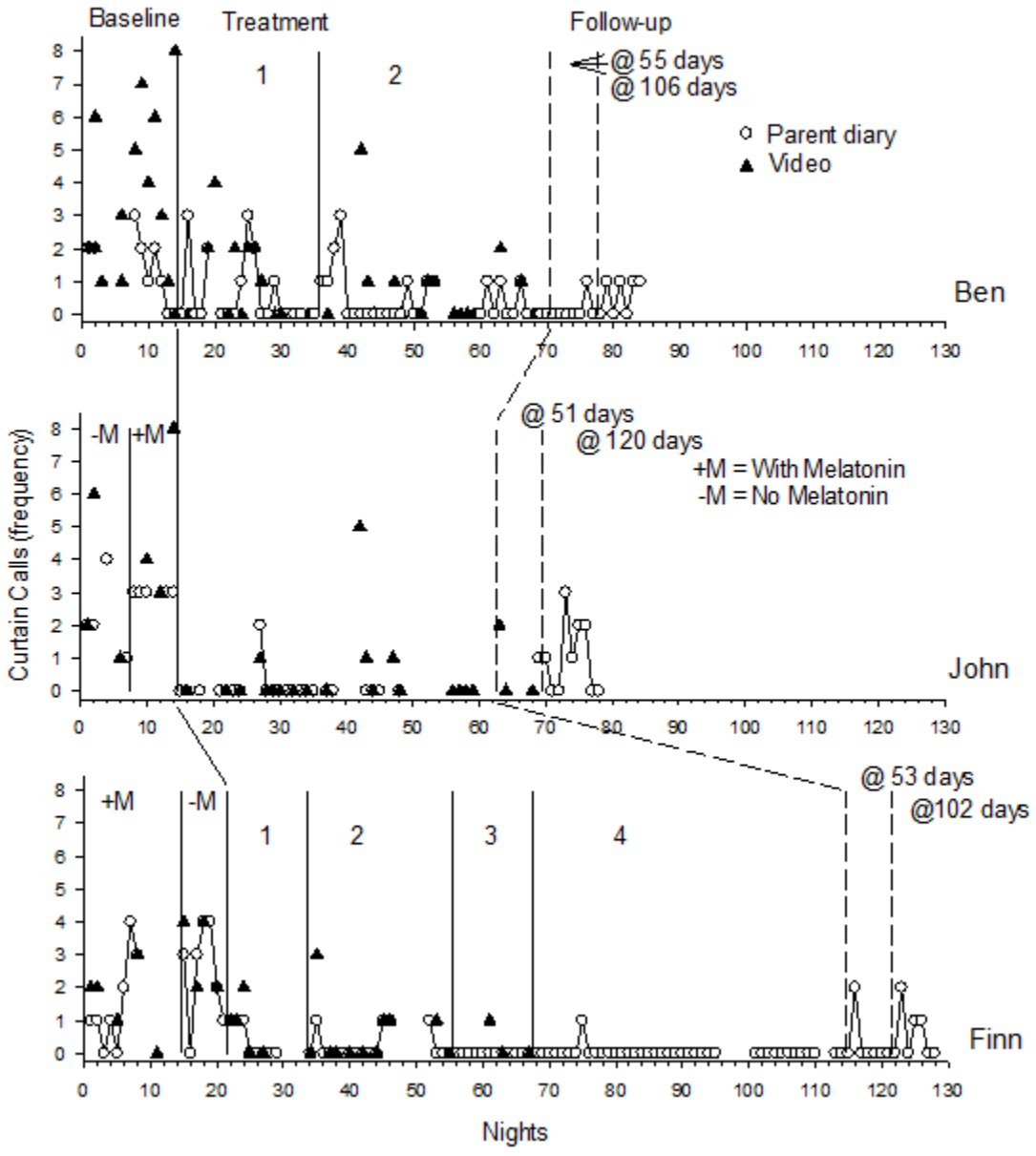

Figure 5.1. Frequency of curtain calls per night across study phases for Ben, John, and Finn. +M = baseline with melatonin; -M = baseline without melatonin. Refer to Table 5.4 for a description of Ben and Finn's treatment phases.

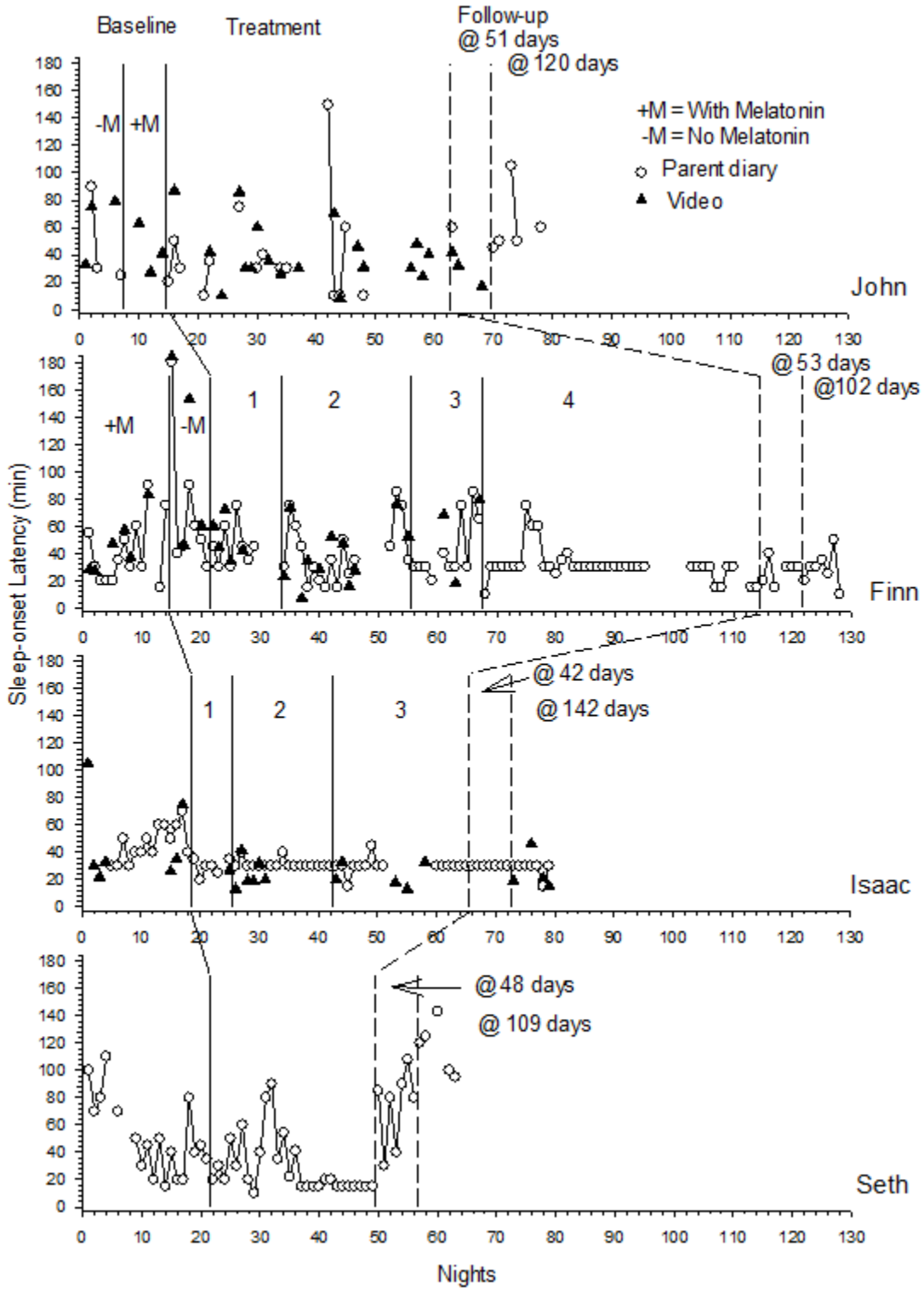

Figure 5.2. Duration of sleep onset latency per night across study phases for John, Isaac, Seth, and Finn. +M = baseline with melatonin; -M = baseline without melatonin. Refer to Table 5.4 for a description of Finn and Isaac's treatment phases.

NWs

There was a moderate to significant reduction in the duration of Blair (PBM_{Video} = 89%) and Ben's (PBM = 96%) respective NWs by the end of treatment. This was maintained at short- (Blair and Ben PBM = 100%) and long-term (Blair and Ben PBM = 100%) follow-up for both participants (see Figure 5.3).

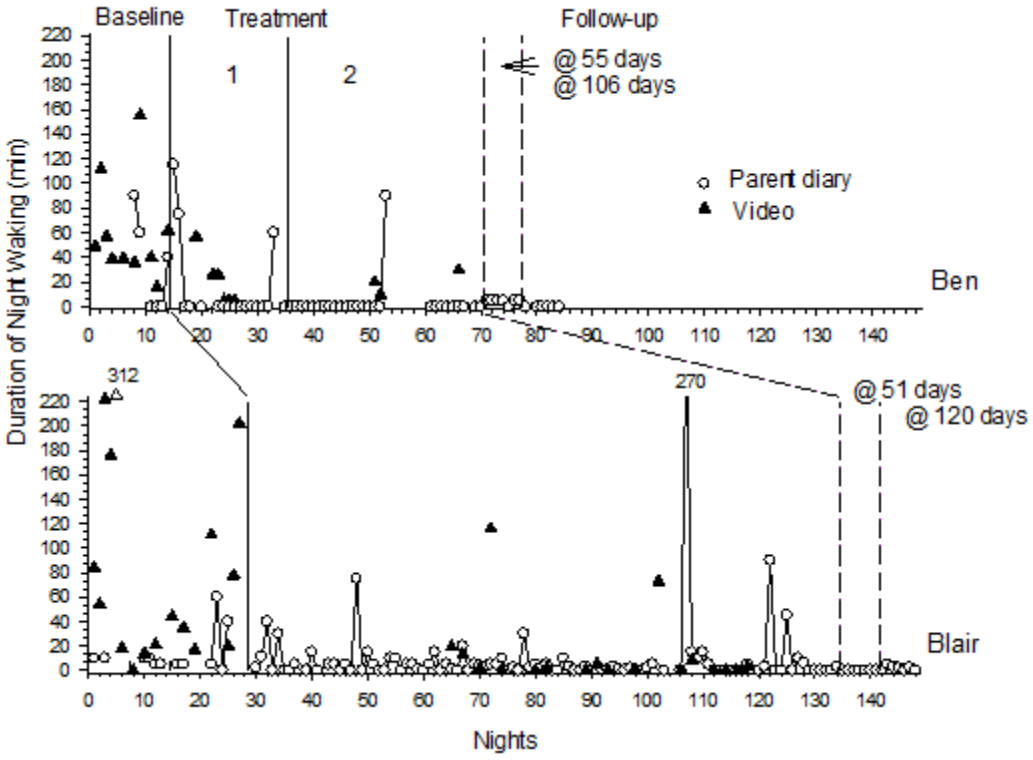

Figure 5.3. Duration of wakings per night across study phases for Ben and Blair. Refer to Table 5.4 for a description of Ben's treatment phases.

Co-sleeping

Both Will and Scott experienced a substantive improvement in the frequency of independent sleep onset at bedtime, with gains maintained at follow-up (see Figure 5.4). Co-sleeping following NW was eliminated during treatment and maintained at follow-up (see Figure 5.5).

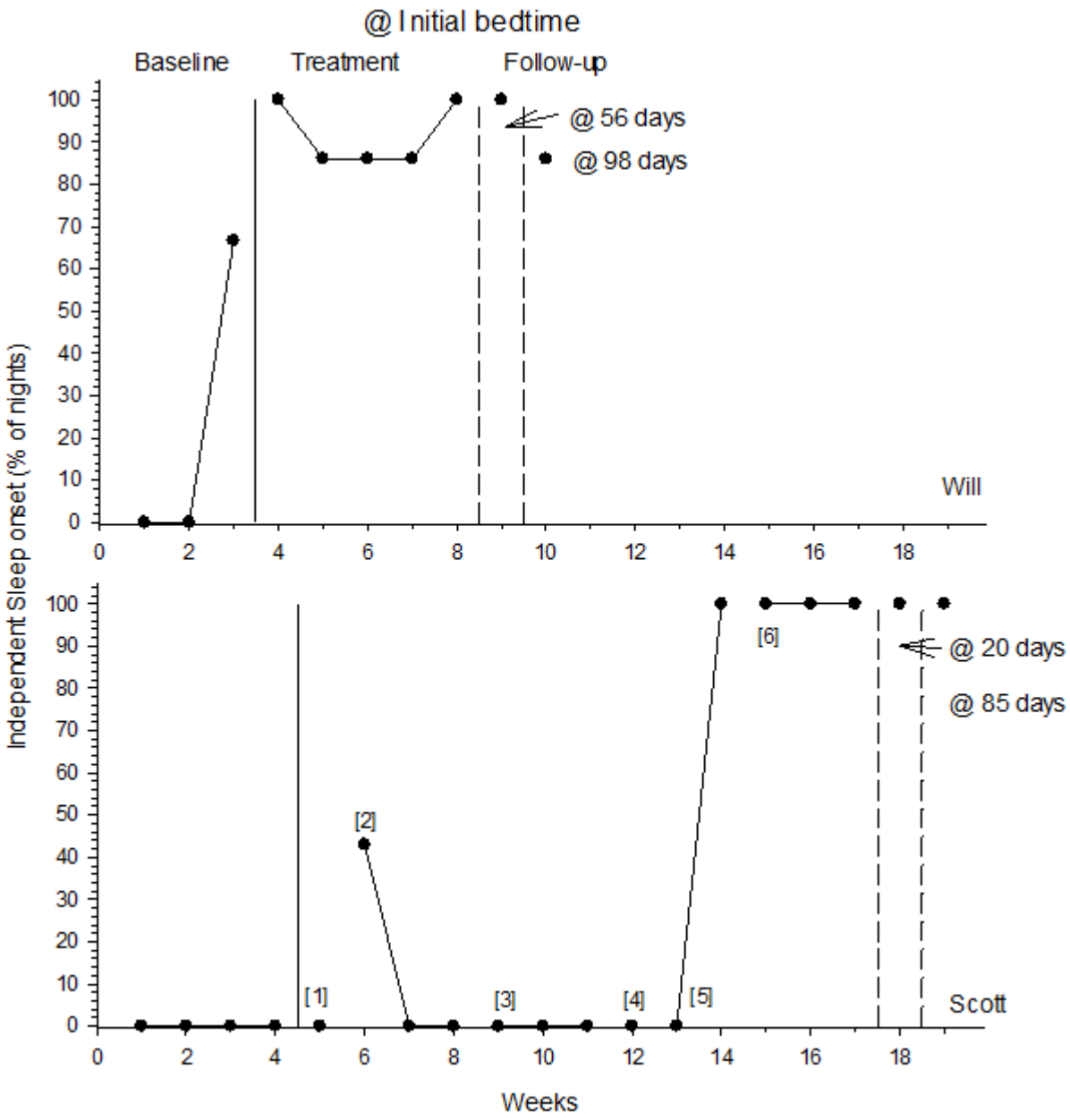

Figure 5.4. Percentage of nights per week Will and Scott fell asleep independently at bedtime. Refer to Table 5.4 for a description of Scott's treatment phases.

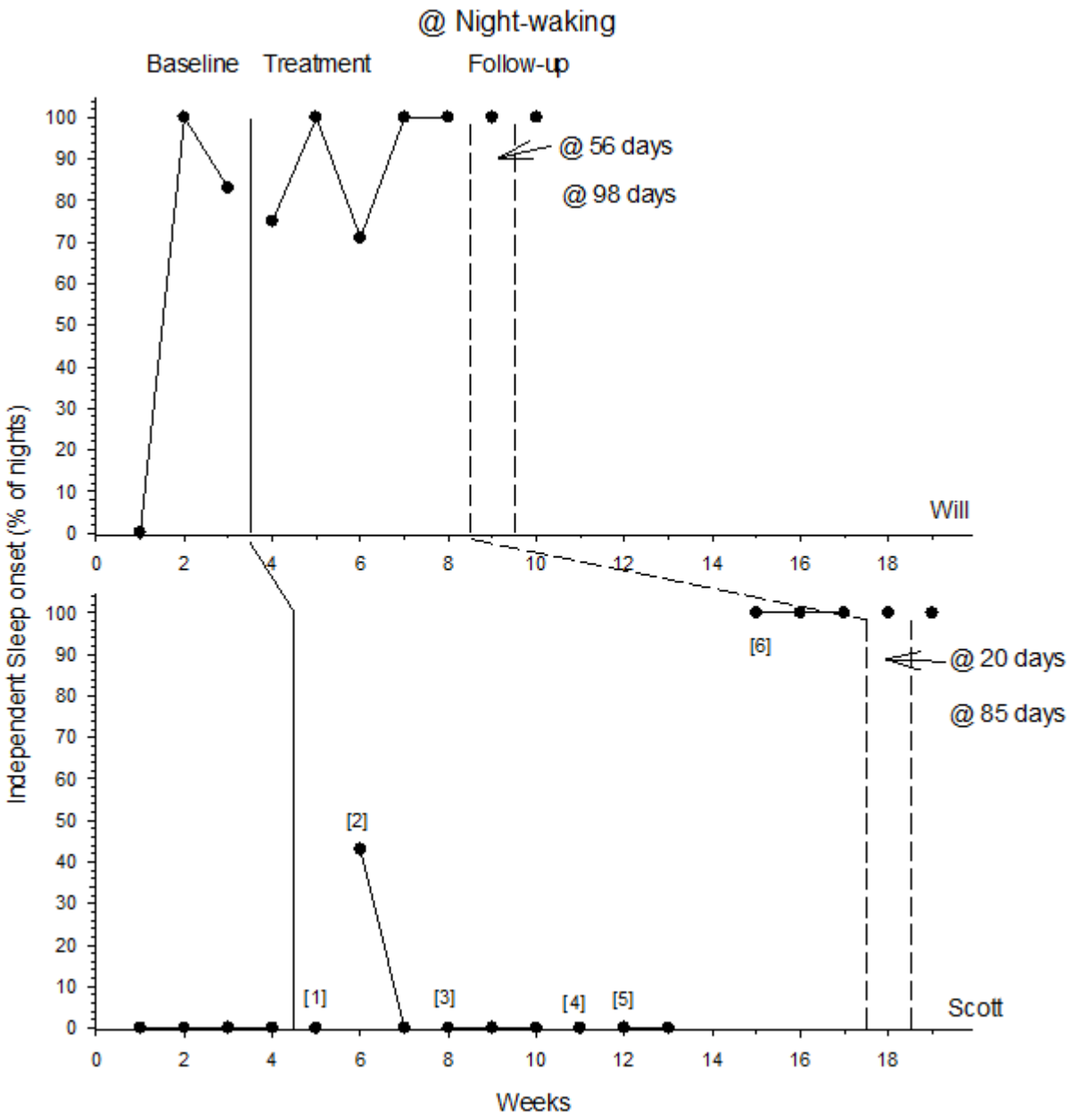

Figure 5.5. Percentage of nights per week Will and Scott reinitiated sleep independently upon waking. Refer to Table 5.4 for a description of Scott's treatment phases.

Motivation

Pre-treatment, most participants indicated their sleep was of moderate to high importance and needed to be addressed, however confidence in their abilities to do so varied (range, 2/10 to 10/10). Participants' intentions to engage in sleep-conducive behaviour increased from pre- to post-treatment (see Table 5.5). On the BIQ they were more likely to endorse items with the statement "I will" (48% vs 65%) post-treatment, and less likely to endorse items demonstrating less commitment, such as "I don't plan to" (26% vs 13%).

Table 5.5. *Participant Endorsement of Intention to Change Sleep Behaviours according to the Behavioural Intentions Questionnaire*

	Pre-treatment	Post-treatment
"I tried to"	17%	22%
"I will"	48%	65%
"I need to"	4%	0%
"I want to"	4%	0%
"I don't plan to"	26%	13%

Participant Social Validity

Participants' interview responses indicated treatment reduced sleep problems for 6/8 participants (e.g., "everything that was wrong with my sleep is now fixed", "I weren't sleeping…was tired and grumpy…now can stay in bed"). Regular contact with a researcher was considered helpful and fun (e.g., "you listened", "cool new experience [talking through Skype]") to some participants, whereas others indicated they would have preferred to spend time engaging in preferred activities. One participant also noted that information and support could have come from a parent instead. Participants enjoyed individualised treatment strategies, including faded bedtime, special time with a parent prior to bed, VSM, reinforcement systems, and bedtime passes. Most participants also reportedly found the sleep checklist and relaxation strategies to be helpful.

Participant ratings on the YPTE yielded an average score of 21 (range, 12–28; see Table 5.6). In general, participants rated treatment as being reasonably enjoyable, fair, and non-time consuming (excluding one participant). Overall, participants considered treatment to be acceptable and moderately to highly effective.

Table 5.6. *Young Person Treatment Evaluation Scores*

Scale	Ben	Finn	John	Isaac	Maximum Score
Effectiveness	6	10	8	6	10
Enjoyableness	1	5	3	3	5
Time	1	5	3	3	5
Fairness	1	3	3	5	5
Overall	3	5	5	5	5
Total	12	28	22	22	30

Parent Social Validity

During interviews parents emphasised the importance of including their child in the therapeutic process and felt this was critical to success. All parents reported their child took on more responsibility for their own sleep and one parent noted their child was empowered through their participation. Regular contact between parents and the therapist was thought to contribute to intervention effectiveness as it facilitated frequent problem solving. Sustainable resources (e.g., video model) which could be used in the future to prevent relapse were valued and VSG was considered crucial to understand the extent of their child's sleep disturbance. Many parents commented there were fewer arguments at bedtime, reducing their own stress and fatigue. Additionally, they believed their own sleep had improved as they were no longer being disturbed at night. Further, they reported improvement in their child's activity levels, organisation, performance in extracurricular activities, and emotional regulation. One parent reported it was unrealistic to expect their child to engage in behavioural quietude once in bed (e.g., refraining from reading which was perceived to be calming and sleep-conducive), contributing to inconsistent implementation of the treatment plan. Another parent reported engaging in special time with their child prior to bed was disruptive to the family routine and reduced parental alone-time in the evening.

Parent ratings on the TARF-R yielded an average score of 103 (range, 79–114; see Table 5.7). Overall, parent responses indicated the interventions were effective, reasonable, easy to understand, and low cost. As reflected in post-treatment interviews, some parents noted there were negative side effects to the intervention (e.g., child resistance) and that treatment was at times, time consuming and/or disruptive.

Table 5.7. *Treatment Acceptability Rating Form-Revised Scores*

	Seth	Finn		Scott		Ben	John	Isaac		Maximum Score
Scale	Mother	Mother	Father	Mother	Father	Mother	Mother	Mother	Father	
Effectiveness	14	21	17.5	21	21	16	19	17	19	21
Reasonableness	15	21	18	21	20	18	21	20	20	21
Willingness	17	21	19	18	21	14	20	20	20	21
Cost	14	14	12	14	12	11	14	14	14	14
Negative side-effect	9	21	19.5	17	21	13	10	19	21	21
Disruption/time	10	13	17	21	19	14	10	21	17	21
Problem severity*	11	10	10	10	11	10	3	9	9	14
Understanding of treatment*	6	7	6	7	6	6	7	6	6	7
Total acceptability	79	111	103	112	114	86	94	112	113	119

Note. *Not included in total acceptability score

Discussion

The purpose of this study was to evaluate the effectiveness and social validity of FBA-informed young person-implemented sleep interventions, which included parent assistance as needed. Overall, intervention implementation coincided with moderate to significant improvement in all target sleep variables across participants. Treatment effects were maintained across participants for duration of NWs, and independent sleeping practices.

Improvements in CCs were not maintained for one out of three participants, and one participant experienced a deterioration in SOL at long-term follow-up.

In general, participants and their parents considered treatment to be effective and acceptable. Participants particularly enjoyed reinforcement systems and valued individualised treatment components. Overall, parents felt their child's active involvement in the intervention contributed to its effectiveness. Interestingly, parent-implemented treatment components were considered the most disruptive elements (e.g., limiting access to books post-bedtime, spending more time with their child prior to bedtime). It was important to young people and their parents that treatment was not too time intensive.

Critically, the current findings provide evidence of the effectiveness of FBA-informed interventions for older children and adolescents. Further, the results support the use of young person-implemented behavioural sleep interventions. Not only were such components preferred, but they were sufficient to treat sleep disturbance in two cases. Additionally, this study reveals numerous methods by which young people can be actively included in the assessment and treatment process, such as through use of individualised self-report sleep diaries, creation of a child social validity measure, and providing psychoeducation and teaching skills directly to the young person.

The results of this study extend previous research by illustrating the feasibility of actively including young people with ASD in the implementation of their behavioural sleep interventions (Loring et al., 2016; McCrae et al., 2019; Souders et al., 2017; van Deurs et al., 2019), in turn, minimising parent-led input. Will and Isaac experienced a significant reduction in sleep disturbance via young person-implemented treatment components, without parent input. For Will, this was achieved following just one session of psychoeducation and relaxation instruction, in addition to these techniques Isaac received support to bring his sleep under appropriate stimulus control. Participant treatment components, with the addition of parent-implemented reinforcement, were sufficient to address Finn and Ben's sleep problems. To address the function of Seth, John, and Blair's sleep-interfering behaviour, antecedent- (e.g., special parent-child time prior to bed) and consequence-based interventions involving parent input were necessary. Systematic fading of parent presence was also required to establish Scott's independent sleeping practices.

Despite parent behaviour (e.g., inadvertent reinforcement of sleep-interfering behaviour) being implicated in all participants' sleep disturbance, it was not always necessary to directly address this through parent-implemented treatment. Reinforcement contingencies which maintained sleep-interfering behaviour may have been altered by participant behaviour change. For example, Finn rarely left his room post-bedtime during intervention, limiting capacity for parent-child interaction and inhibiting social reinforcement for sleep-interfering behaviour. This may have contributed to a shift in parental attributions and behaviour (e.g., feeling confident their child could manage independently, and responding in a less intrusive manner).

Importantly, the effectiveness of young person-implemented treatment components meant sleep disturbance was addressed via application of less restrictive methods for most participants (e.g., psychoeducation, skills training, reinforcement). Notably, CCs, which by their very nature involve social reinforcement, were treated without using extinction-based techniques. Although, a modified extinction procedure (systematic fading of parental presence) was used to increase Scott's independent sleeping practices, no resistance occurred; perhaps due to the gradual nature of this intervention and the addition of participant treatment components. Scott's parents reported regular contact and encouragement from the therapist provided him a sense of "choice" and "control over it", which increased his confidence that he could successfully achieve each step.

The effectiveness of young person-implemented treatment components may have been mediated in part by participant communicative abilities, behavioural function, and intrinsic motivation. Will and Isaac had the highest communicative abilities and required the least parent input. Conversely, Blair and Scott had the lowest communicative abilities and required the most parent input. Intrinsic motivation may be linked to communicative abilities, since participants who had a stronger understanding of the implications of sleep disturbance may have been more intrinsically motivated to address their sleep disturbance. Interestingly, Blair ceased device use during NWs following psychoeducation, and requested his parents unplug his devices post-bedtime. However, this was not sufficient to address his sleep disturbance as it was also necessary to modify access to social reinforcement, suggesting this was a primary function of his sleep-interfering behaviour. Participants who rated sleep as being of high importance (Will, Isaac, Finn, and Ben) tended to require less parent input, regardless of how confident they were their sleep could be improved. Will and Isaac had high

intrinsic motivation to address their sleep disturbance. All remaining participants required rewards to encourage engagement in sleep-conducive behaviour, with Blair, Seth, and Scott requiring implementation of extinction procedures to address sleep-interfering behaviour.

Rates of participant and parent treatment fidelity varied considerably between families, limiting capacity for generalization across the sample. Nevertheless, overall participant and parent treatment fidelity tended to reduce during follow-up, regardless of participants' intentions to continue to engage in sleep conducive behaviour (i.e., endorsement of "I will" on BIQ target behaviours). In general, maintenance of treatment effects was dependent on continued fidelity by the primary intervention agent, whether this was the young person or parent. Seth experienced the biggest reduction in fidelity, which likely contributed to the significant deterioration in SOL at follow-up. The intellectual abilities of the participants may have compromised their capacity to self-regulate their behaviour over an extended period, particularly without extrinsic reinforcement and accountability. In accordance with therapist instruction, reward systems were faded by the end of treatment for some participants, however during follow-up some parents stopped delivering reinforcement by their own initiation, a lack of reinforcement for sleep-conducive behaviour may have undermined participant motivation. At times, parents may alter treatment to make it easier to implement, compromising treatment integrity (Healey, France, & Blampied, 2009). Unsurprisingly, Seth's treatment fidelity reduced, and John's remained low at follow-up after both reporting they did not intend to continue engaging in unfavourable treatment components (consistent sleep/wake schedule, no reading post-bedtime) post-treatment.

While not a key focus of this study, the results revealed interesting findings relating to the relative effects of melatonin versus a behavioural intervention for sleep. Critically, as this analysis was only carried out with two participants, the ability to make inferences is limited and the findings should be interpreted with caution. Both Finn and John demonstrated positive effects of melatonin on SOL, however, these effects were matched by behavioural intervention, which in turn outweighed the effects of melatonin alone on CCs. Results suggest behavioural intervention and behavioural intervention + melatonin conditions can produce similar effects for SOL as melatonin alone, but superior effects for CCs. Similarly, Cortesi et al. (2012) found behavioural therapy + melatonin was superior to melatonin alone to improve sleep disturbance in children with ASD. In contrast to the current study, they found behavioural therapy alone was not superior to melatonin alone. This difference could

potentially be explained by the increased effectiveness of FBA-informed interventions. While melatonin can address physiological contributions to sleep disturbance (e.g., reduced sleep pressure), it may not resolve sleep problems maintained by environmental contingencies (e.g., access to social attention). These results emphasise the importance of conducting a thorough functional assessment to ensure key factors underlying sleep disturbance are addressed.

Of note, the current study also highlights the incongruence between widely used sleep measures. Comparison between self-report sleep diary data and video observations illustrated that Isaac tended to overestimate SOL, as is common among individuals with insomnia (Tang & Harvey, 2005). Conversely, VSG showed parents tended to underestimate their child's sleep disturbance. Based on sleep diaries alone Blair and Ben's target variables would not be considered problematic. IOA between parent-reported sleep diaries and video observations for detectable sleep phenomena across participants and target variables was 80%, however, overall IOA between parent-reported sleep diaries and video observations was 65%, indicating the significant difference between phenomena detectable via video observation versus parent observation. This finding is consistent with Bauer and Blunden's (2008) review of data consistency between sleep diaries and questionnaires and PSG and actigraphy for infants through to adolescents. Parents and adolescents could correctly identify observable variables, such as bedtime and final wake time, but had difficulty identifying SOL and the frequency and duration of NWs (Bauer & Blunden, 2008). In contrast to van Deurs et al. (2019), when CCs occurred in high frequency, it was difficult for parents to note each instance and they tended to be underestimated. These results emphasise the importance of using a range of measures of sleep in research and clinical practice to accurately understand sleep within paediatric populations. Sleep assessments using measures that tap reliably and validly into different dimensions of sleep and associated circumstances are warranted given they each capture unique phenomena (e.g., heart rate, movement, psychological states) and do not always produce consistent results between measures (Baddam, Canapari, van Noordt, & Crowley, 2018).

Several methodological issues need to be considered. Firstly, video observations were not collected during follow-up for all participants and parent report cannot necessarily be relied on. Therefore, maintenance of treatment effects should be interpreted with caution. Secondly, the design of the current study inhibited evaluation of specific treatment

components. Future research might investigate which components are necessary to appropriately address the function of sleep-interfering behaviour in a minimally sufficient manner to enhance social validity. Thirdly, this study was conducted with verbal young people with ASD and its findings may not apply to non-verbal individuals. Finally, lack of a comparison group (who received parent treatment components only) in the current study and the absence of participant social validity data within previous sleep intervention research, means it is unknown whether the current treatment was more or less acceptable than traditional parent-implemented sleep interventions. Overall, results extend previous findings (van Deurs et al., 2019) and provide additional evidence for the use of individualised, FBA-informed interventions involving input from the young person with ASD and their parents to address a range of sleep problems.

Chapter 6: Study 3

Sequential Implementation of FBA-Informed Treatment Components for Sleep Disturbance in Autism: a Case Study[3]

The Ministries of Health and Education (2016) estimate 1 in 100 people in NZ have ASD; a neurodevelopmental disorder characterised by difficulties with social communication and restricted/repetitive thoughts and behaviour patterns. In addition to these two core features, the neurological functioning of people on the autism spectrum can compromise their speech and language skills, sensory responsivity, executive functioning (e.g., emotional and behavioural regulation, attention), and psychological wellbeing (e.g., anxiety). Sleep disturbance is also a significant clinical problem faced by many people on the autism spectrum. Rates of sleep disturbance in children and adolescents with ASD range from 50 to 80% in studies conducted in the USA and Canada (Couturier et al., 2005; Krakowiak et al., 2008; Malow, Katz et al., 2016; Souders et al., 2009). Difficulty initiating and maintaining sleep, as evidenced by prolonged sleep latency, shorter sleep duration, NWs, and reduced SE are the most common sleep problems experienced by young people with ASD (Herrmann, 2016). These issues tend to persist throughout childhood and adolescence if untreated (Sivertsen et al., 2012), although the phenomenology changes with age (Goldman et al., 2012). Parents of children with ASD report higher rates of bedtime resistance, NWs, parasomnias and sleep anxiety, whereas parents of adolescents with ASD report higher rates of delayed sleep onset, shorter sleep duration, and daytime sleepiness (Goldman et al., 2012).

Sleep deprivation exacerbates the difficulties already faced by young people on the autism spectrum, further compromising their cognitive and adaptive functioning, behaviour, and emotional wellbeing (Nadeau et al., 2015; Sikora et al., 2012; Park et al., 2012). For example, mild ASD severity is typically associated with lower levels of behaviour problems than higher ASD severity, however, young people with ASD and sleep disturbance are likely to exhibit clinical levels of problem behaviour, regardless of ASD severity (Lindor et al., 2019). Further, the effects of sleep disturbance are not isolated to the individual but can also affect family functioning. Parents of young people with ASD and sleep problems report

[3] An article based on this study has been published in Behavioral Sleep Medicine: van Deurs, J. R., McLay, L. K., France, K. G., & Blampied, N. M. (2020). Sequential implementation of functional behavioural assessment-informed treatment components for sleep disturbance in autism: A case study. *Behavioral Sleep Medicine.* Advance online publication. doi:10.1080/15402002.2020.1758701 (See Appendix W).

higher stress and poorer mental health than parents of young people with ASD without sleep problems (Martin, Papadopoulos, Chellew, Rinehart, & Sciberras, 2019).

Bidirectional interactions between physiological (e.g., dysregulated melatonin), behavioural (e.g., classical and operant conditioning of sleep-interfering stimuli), and psychological (e.g., comorbid psychopathologies) factors contribute to the aetiology of sleep problems in people with ASD. While pharmacological sleep interventions (e.g., prescribed melatonin) can address underlying physiological contributions, they neglect other factors precipitating and maintaining the sleep problem. A growing evidence base supports the use of parent-implemented behavioural interventions for the treatment of sleep disturbance in children with ASD (Carnett, Hansen, McLay, Neely, & Lang, 2019). There is also emerging evidence for the use of combined young-person- and parent-implemented cognitive behavioural interventions for older children and adolescents with ASD (Loring et al., 2016; McCrae et al., 2019; van Deurs et al., 2019).

FBA is commonly used to identify antecedent and consequence variables maintaining problem behaviour. A number of studies highlight the importance of using FBA to inform sleep interventions for young people with ASD (e.g., Jin et al., 2013; McLay, France, Blampied et al., 2019; McLay, France, Knight et al., 2019) and indicate that similar behavioural topographies (e.g., leaving the bedroom post-bedtime) do not necessarily serve the same function, nor warrant similar treatment. For one child, sleep-interfering behaviour may be maintained by parent attention, and for another via access to tangible items. This information is critical to the development of targeted, effective interventions.

A recent review of behavioural interventions for sleep disturbance in children with ASD revealed all existing studies have consisted of multiple components (Carnett et al., 2019). Consequently, the specific mechanisms of behaviour change are unknown. As the effects of each component have not been evaluated independently from one another, it is unclear which strategies are necessary and minimally sufficient to produce change. Treatment is less likely to be adhered to when it is disruptive to family routine, time intensive, complex, or evokes distress. This is concerning given comprehensive FBA-informed treatment packages can be challenging to deliver and involve extensive time commitment. The minimal sufficiency principle emphasises the implementation of treatment components which balance effectiveness with ease of delivery (Sanders et al., 2014). Similarly, in accordance with the least restrictive alternative principle, the least aversive and intrusive methods capable of

producing significant therapeutic change should be used (Kazdin, 1984). The identification of least restrictive and minimally sufficient methods to address the function of sleep-interfering behaviour may be critical to facilitating treatment fidelity and maintenance of behaviour change.

A range of objective and subjective measures can be used to assess sleep, with each capturing unique phenomena within the same construct. Sleep diaries gather continuous data on sleep patterns for a period of time, whereas questionnaires require informants to make retrospective approximations about sleep patterns over extended time periods and may be at increased risk of recall bias. Research indicates discrepancies between information obtained from objective and subjective measures. For example, parents are simply not able to detect covert wakings whereby their child remains quiet in bed, evident via video observation. Parents and adolescents are reliable informants of overt variables, such as bedtime and waketime, but they have difficulty evaluating covert variables, such as duration of sleep onset or NWs (Bauer & Blunden, 2008). VSG has been used rarely within the ASD and sleep literature. Additionally, self-report sleep diaries (commonly completed by typically developing young people) have been scarcely used by young people with ASD. Further, no single study within the ASD and sleep literature has compared information obtained from parent-report sleep diaries, self-report sleep diaries, VSG, and questionnaire outcomes.

The aims of the present study were to (a) sequentially administer, minimally intrusive, FBA-informed treatment components in an attempt to resolve sleep disturbance in a child with ASD; (b) examine parent- and child-reported social validity of treatment approaches; and (c) compare information gathered via parent-report sleep diaries, self-report sleep diaries, VSG, and questionnaires.

Method

Participant

Eve (pseudonym) was a 9-year-old girl with autism and selective mutism. The Communication sub-domains of the Vineland-3 revealed Eve had below average receptive and expressive language abilities, equivalent to a 2 year, 9-month-old child, and a 3 year, 11-month-old child respectively. She had high reading and writing skills, equivalent to an 8 year, 6-month-old child. Analysis of Vineland-3 items and clinical assessment revealed Eve could ask and answer questions that involved “when” and “why, she could sometimes describe

everyday events in detail, write or draw instructions for others, fill out forms with more than two pages, and attend to and understand a 30-minute informational talk. Eve was referred to the study by her parents. Eve's parents reported she took an extended period of time to initiate sleep at bedtime (60 - 120 min), and engaged in multiple CCs. They reported Eve woke every night, occasionally (up to three times a week) for several hours. Eve received 2.5mg of slow-release melatonin nightly.

Design

This study employed an AB single-subject design whereby the intervention phase consisted of three sub-phases with cumulative addition of treatment elements, namely white noise; white noise and relaxation instruction; and white noise, relaxation instruction, and stimulus control. Treatment effects were replicated across five target sleep variables, including CCs, SOL, NWs, total sleep duration, and SE.

Setting

Pre- and post-treatment assessment interviews and treatment planning discussions were conducted in the family home. Treatment was implemented within the home by Eve and her parents with support from the researcher. During treatment, communication with Eve's parents was conducted in person, or via telephone, and email. As Eve experienced high anxiety speaking on the telephone, the researcher communicated with her face-to-face and via letters using language appropriate for her reading level.

Measures

Clinical interviews. A semi-structured interview was conducted separately, with Eve and her mother. Information was gathered regarding Eve's interests and strengths; developmental history; current and historic sleep concerns; factors contributing to sleep disturbance (e.g., Eve's thoughts and emotions at bedtime); motivation to improve sleep; and family context. To facilitate communication with Eve the interview included: visual stimuli (e.g., sleep cartoons); drawing and written activities; incorporation of Eve's special interests; metaphors (e.g., magic wand to address sleep disturbance); and open, closed, and multiple-choice question format. Eve was given extended time to answer questions and typically did so within 10 to 20s.

SATT. The SATT was incorporated in the clinical interview with Eve's mother. The SATT is a semi-structured interview used to identify factors underlying children's sleep disturbance.

QABF. The QABF was administered to Eve's mother following the clinical interview (enabling further explanation of items as needed). The QABF is a 25-item rating scale used to establish the function (e.g., social attention, escape, tangible reinforcement) of a target behaviour.

MASC-2. The MASC-2 was completed by Eve and her parents during assessment to measure existing anxiety levels. The MASC-2 is a 50-item multi-informant questionnaire designed to assess anxiety symptoms (e.g., Social Anxiety, Separation Anxiety, Physical Symptoms) experienced by people aged 8 to 19 years. The number of elevated scores across Anxiety Scales yield an Anxiety Probability score, classified as Low, Borderline, High, or Very High.

CSHQ. The CSHQ was completed by Eve's parents during the assessment and maintenance phase to evaluate change in parent-reported sleep disturbance. The CSHQ is a widely used parent-report questionnaire for assessing school-aged children's sleep patterns according to the frequency of specific behaviours across eight sleep domains (Bedtime Resistance; Sleep Onset Delay; Sleep Duration; Sleep Anxiety; Night Wakings; Parasomnias; Sleep Disordered Breathing; and Daytime Sleepiness) within a typical week. Higher scores are indicative of poorer sleep, and scores ≥ 41 are indicative of clinically significant sleep disturbance (Owens, Spirito, & McGuinn, 2000). The CSHQ has satisfactory internal consistency (0.68 to 0.78) and test-retest reliability (0.62 to 0.79; Owens, Spirito, & McGuinn, 2000).

The Sleep Self-Report (SSR; Owens, Spirito, McGuinn & Nobile, 2000). The SSR was completed by Eve during assessment and upon entering maintenance phase as a measure of child-reported sleep pre- and post-treatment. The SSR is a 26-item self-report questionnaire for children 7 to 12 years of age which corresponds with the CSHQ subscales (Owens, Spirito, McGuinn & Nobile, 2000). Eve rated the frequency of specific sleep behaviours engaged in over the past week on a 3-point Likert scale (Owens, Spirito, McGuinn & Nobile, 2000). SSR scores have a possible range of 23 to 69 with higher scores indicative of worse sleep. The SSR has adequate discriminative validity and test-retest

reliability (0.76 - 0.88; Orgilés, Owens, Espada, Piqueras, & Carballo, 2013; Owens, Spirito, McGuinn & Nobile, 2000; Steur et al., 2019).

Sleep diaries. Eve's parents completed daily sleep diaries during each phase of the study. Recorded information included the (a) frequency and duration of daytime sleep; (b) duration of SOL; (c) frequency of CCs; (d) frequency and duration of NWs; and (e) time of morning waking. Eve's sleep setting, behaviour during CCs and NWs, and parent responses to this behaviour were also noted. Eve was also asked to complete daily sleep diaries which included the addition of the type and intensity of emotions experienced before bedtime.

VSG. A D-Link HD Cloud Camera was used to directly observe and measure Eve's sleep during each study phase. The camera was placed at the end of Eve's bed and was set to turn on at her typical bedtime and switch off at her typical rise time. Information obtained from the video included the same information reported in sleep diaries, plus the addition of topographies of awake behaviour post-bedtime (e.g., vocalisations, play) and topographies of sleep behaviour (e.g., sleep position, eye movement, limb movement). The following operational definitions were used to code video (a) asleep, lying down with minimal non-discrete movement for ≥5 min, and no indication of wakefulness; and (b) awake, the presence of any sleep-interfering behaviour, eyes open, or frequent physical movement (Jin et al., 2013). Video footage was coded by a researcher blind to parent sleep diary recordings and enabled objective monitoring of Eve's progress and the collection of IOA data.

IOA. Agreement between parent- and self-report diary data and video observations, was calculated. Sleep phenomena which parents could not be expected to detect (e.g., covert awakenings in which Eve remained quiet in her bed) were omitted from IOA calculations. IOA for CC frequency was calculated on occasions whereby bids for attention were detectable by video observers (e.g., clear calling out). Measures of duration (e.g., SOL) and sleep/wake times were considered in agreement if they were ± 15 min. Percent agreement for target behaviours was calculated using the equation [Agreement/ (Agreement + Disagreement)] × 100. IOA data were collected for 35% of nights across all study phases.

Treatment fidelity. Treatment fidelity was assessed on 88% of nights across intervention and follow-up phases by comparing measurable events recorded in contact notes, video footage, and sleep diaries with the prescribed treatment protocol. An aggregate

treatment fidelity score was calculated using the formula (Completed tasks/ Total tasks) × 100.

Social validity. Eve completed the YPTE post-treatment to assess her perception of the intervention. The YPTE, developed by the authors based on the Child Evaluation Inventory (CEI; Kazdin, 1984), consists of six items assessing effectiveness, enjoyability, fairness, time required, and overall perception, using a 3-point Likert scale (e.g., 1 = Not at all helpful; 2 = OK; and 3 = Very helpful). Ratings are summed to provide a total acceptability score. Post-treatment, Eve's parents completed the TARF-R, a 20-item questionnaire based on the adult version of the CEI. Ratings on six subscales (Effectiveness; Reasonableness; Willingness; Cost; Negative side-effects; Disruption/time) are summed to provide a total treatment acceptability score. Post-treatment interviews were also conducted with Eve and her parents individually to further evaluate social validity and gather qualitative information regarding treatment effects.

Procedure

FBA. Information obtained from the SATT, the QABF, sleep diaries, and analysis of video footage were used to conduct the FBA and synthesised in an FBA-informed case conceptualisation which then guided treatment planning (Blampied, 2013a). FBA indicated that many antecedent and consequence variables were contributing to Eve's sleep disturbance. Table 6.1 describes the factors hypothesised to precipitate and maintain each of Eve's sleep difficulties as well as their function.

Assessment revealed Eve's bedtime varied between 8:00 to 9:00pm. Once in bed, Eve played games on an iPad, listened to podcasts, or read using an e-reader until she fell asleep. Her parents tended to bid her goodnight and switch off the light at variable times during this period, following which Eve continued to engage in sleep-interfering activities. Eve's bedroom was situated close to the main living areas and she kept her bedroom door open at night; she complained the television volume was too loud post-bedtime. Parent ratings on the MASC-2 yielded Total Anxiety scores within the Very Elevated range and indicated a Very High Probability of an anxiety disorder. This was corroborated by self-report ratings on the MASC-2. Additionally, Eve reported experiencing frequent and intense worry about her parent's wellbeing and consequently regularly listened to and monitored her parents' conversations post-bedtime, interjecting at times. Further, she typically called out to her

parents multiple times to request food or drink, and discuss other worries. Her parents responded inconsistently, providing comfort/reassurance and/or requested items, or reprimanding the behaviour. Eve usually woke multiple times per night for no identifiable reason or in response to her baby sibling crying, then experienced difficulty reinitiating sleep. These wakings generally resulted in her using her e-reader. On some occasions Eve did not return to sleep at all.

Table 6.1. *Factors Precipitating and Maintaining Sleep Disturbance, Hypothesised Function, and Treatment Components*

	Curtain calls	Delayed Sleep Onset	Frequent and extended night wakings
Factors thought to be precipitating and/or maintaining behaviour	Lack of physiological sleep pressure; parent responses to curtain calls; cognitive and physiological hyperarousal	Lack of physiological sleep pressure; lack of discriminative stimuli for sleep; salient discriminative stimuli for sleep-competing behaviour (e-reader); loud external noises; cognitive and physiological hyperarousal	Lack of physiological sleep pressure; lack of discriminative stimuli for sleep; salient discriminative stimuli for sleep-competing behaviour (e-reader); loud external noises; cognitive and physiological hyperarousal
Hypothesised function	Social attention Access to tangibles Escape (from distressing cognitions)	Social attention Access to tangibles	Access to tangibles

Antecedent variables hypothesised to be implicated in Eve's sleep disturbance included lack of physiological sleep pressure (abolishing operations for sleep), lack of consistent discriminative stimuli for sleep, salient discriminative stimuli for sleep-competing behaviour (e-reader); external noise; and cognitive and physiological arousal. Sleep-interfering behaviour was thought to be reinforced by social attention, access to tangibles (e-reader), and escape from distressing cognitions.

Baseline. Baseline commenced following completion of the FBA. Eve was randomly assigned a baseline length of four weeks. Her family were asked to maintain existing sleep habits during this phase.

Intervention. Intervention commenced immediately following baseline and consisted of three phases. FBA-informed intervention components were implemented sequentially across these phases until Eve's sleep disturbance resolved. Up to five intervention phases were planned; treatment components were selected according to those which were hypothesised to most appropriately address the function of the behaviour and ordered from the least to most restrictive and time intensive. Treatment proceeded to the next phase if clinically substantive improvement across target sleep variables had not occurred within seven days. Eve entered maintenance phase (continuation of the final treatment protocol without therapist input or data collection) when there was a clinically substantive reduction in target behaviours lasting at least 14 days.

Given Eve's difficulty engaging with new people, her parents were responsible for providing psychoeducation and introducing her to therapeutic techniques, following coaching from the researcher. They were contacted daily to weekly by the researcher to facilitate effective treatment delivery and monitor fidelity. Eve had therapist contact at one to two weekly intervals via letters and certificates of achievement.

Treatment phase 1: White noise (night 30 to 40). The first phase of treatment involved playing two preferred sounds, selected by Eve (cat purring and fire crackling) from the Rain Rain application (https://www.rainrainapp.com/) from bedtime to morning wake time, and her door was also closed to mask external noises (e.g., sibling crying, television). These strategies inhibited excessive reassurance seeking as Eve could no longer hear, monitor, and respond to adult conversation from her bed (Eve's parents were not given any instructions regarding how to respond to CCs should they occur). Further, the white noise may have acted as a salient proximate discriminative stimulus for sleep. Eve and her parents chose a volume which masked external noise while remaining at a comfortable level.

Phase 2: White noise and relaxation instruction (night 41 to 51). On night 41, Eve was taught relaxation strategies (diaphragmatic breathing and PMR) to provide her with skills to independently alleviate hyperarousal and facilitate independent initiation of sleep at bedtime and during wakings. She was taught the relaxation strategies during one in-home session with the researcher who used modelling and incorporated Eve's soft toys in the instruction, aided by a short story which included pictures of Eve's favourite animal (cat) completing the exercises (see Appendix S). As well as continuing to use white noise, Eve's

parents were instructed to read the book with Eve each night at bedtime and guide her through the relaxation exercises.

Phase 3: White noise, relaxation instruction, and stimulus control (night 52 to 72). During the third intervention phase Eve's sleep was brought further under appropriate stimulus control; both interoceptive (e.g., tiredness) and exteroceptive (e.g., consistent bedtime) discriminative stimuli for sleep were strengthened, and dependencies non-conducive to sleep were eliminated. Bedtime fading and a consistent sleep/wake schedule increased motivating operations for sleep and ensured Eve had sufficient homeostatic sleep pressure to initiate sleep quickly once in bed. She was instructed to use her bed for sleep only and e-reader use restricted to when she was seated at her desk up until bedtime. Eve's parents instructed her to stop using the e-reader at a consistent time each night and bid her goodnight once she was in bed, providing clear sleep cues (once again Eve's parents were given no instruction regarding how to respond to any CCs). These arrangements functioned to make the bed a discriminative stimulus for sleep. To reinforce Eve for no longer using her e-reader in bed, she was allowed to stay up with her parents in the lounge and use her e-reader until bedtime during the first week of Phase 3.

Phase 4: White noise, relaxation instruction, stimulus control, and reinforcement. The planned fourth intervention phase consisted of parent-delivered positive reinforcement (e.g., a small tangible reward) contingent on Eve's engagement in sleep-conducive behavior (e.g., remaining in bed post-bedtime). Eve's parents would not have received instruction regarding their responses to any CC's.

Phase 5: White noise, relaxation instruction, stimulus control, reinforcement, and unmodified extinction. In order to address the attentional component hypothesized to underlie Eve's sleep disturbance, the planned fifth intervention phase involved eliminating inadvertent social reinforcement for sleep-interfering behavior. Eve's parents would have been instructed not to attend to any bedtime disruptions (e.g., calling out). If Eve left her bedroom, her parents would have been required to return her to bed with minimal engagement.

Follow-up. Follow-up video and sleep diary data were collected for 7 nights at 10 weeks post-treatment.

Data Analyses

Visual analysis (examination of level, trend, and stability) of graphed data supplemented by PBM/PEM (dependent on the therapeutic direction of change; Ma, 2006), was used to evaluate the effect of each treatment phase on target behaviour. PBM/PEM is an effect-size measure for single-case data and can be interpreted as follows: < 70% represents ineffective treatment; 70 to 90% moderate effectiveness; and > 90% high effectiveness (Ma, 2009). Within the current study the baseline median for each target behaviour was compared to the individual data points within each treatment phase.

To indicate whether improvement in target variables was clinically substantive, Eve's baseline and treatment data were compared to developmental norms. Clinical cut-offs are presented in Figures 6.1 to 6.5 to discriminate between appropriate and poor sleep quality. Indicators of good sleep quality in typically developing school-aged children include: < 30 min SOL, ≤ 1 waking, ≤ 20 min of wake-after-sleep-onset (WASO), and ≥85% SE (Ohayon et al., 2017). Conversely, indicators of poor sleep quality for this age group include: > 45 min SOL, ≥ 4 wakings (at least 5 minutes in length), > 40 min of WASO, and < 75% SE (Ohayon et al., 2017). Optimal sleep duration for school-aged children is between 9 to 11 hours, 7 to 8 hours are considered appropriate for some children, and less than 7 hours is not recommended (Hirshkowitz et al., 2015). The current study applied a 7-hour (420 min) total sleep time clinical cut-off. As appropriate levels of CCs have not been described within the literature, the current study defined ≥1 CCs more than 5 times a week as a clinical problem.

Pearson product moment correlations were calculated to measure the strength of the relationship between parent-report sleep diaries, self-report sleep diaries, and video observations. Correlation coefficients of > 0.50 represent a strong relationship, 0.30 to 0.49 a medium relationship, and < 0.30 a small relationship between variables (Cohen, 1988).

Results

Agreement Between Sleep Measures

Mean IOA between parent-report diaries and video was 89% (range, 57-100%) across study phases and between self-report diaries and video was 36% (range, 0-100%) across study phases. IOA values across target variables and study phases are presented in Tables 6.2 and 6.3.

Strong positive correlations were found between data obtained from parent-report sleep diaries and video data for SOL, CCs, and SE (range: r = 0.80-0.82; see Tables 6.4 to 6.8). Smaller correlations were found between parent-report and video data for duration of NWs and total sleep duration (range, r = 0.56-0.57), although they exceeded Cohen's convention for a large relationship. Strong positive correlations were found between self-report sleep diaries and video data for SOL, duration of NWs, and SE (range, r = 0.68-0.85). Small to moderate correlations were found between self-report and video data for CCs and total sleep duration (range, r = 0.21-0.48). Parent-report and self-report diary data were strongly correlated across all target sleep variables (range, r = 0.72-0.87).

The correlations indicate that parent-report, self-report, and video data followed a similar pattern over time (e.g., reduction in sleep problems). However, there were large differences between individual numerical values reported at specific time points across measures.

Table 6.2. *Interobserver Agreement Between Parent-report Sleep Diaries and Video Observations Across Target Variables and Study Phases*

	Baseline	Treatment	Follow-up
Curtain calls	57%	79%	100%
Sleep onset latency	57%	86%	100%
Duration of night wakings	100%	100%	100%
Total sleep time	-	100%	100%

Table 6.3. *Interobserver Agreement Between Self-report Sleep Diaries and Video Observations Across Target Variables and Study Phases*

	Baseline	Treatment	Follow-up
Curtain calls	50%	71%	80%
Sleep onset latency	0%	14%	100%
Duration of night wakings	0%	14%	40%
Total sleep time	0%	0%	60%

Table 6.4. *Pearson Product Moment Correlations Between Sleep Measures for Curtain Calls*

	1	2	3
1. Parent-report sleep diary	-		
2. Self-report sleep diary	0.73	-	
3. Video observations	0.81	0.48	-

Table 6.5. *Pearson Product Moment Correlations Between Sleep Measures for Sleep Onset Latency*

	1	2	3
1. Parent-report sleep diary	-		
2. Self-report sleep diary	0.87	-	
3. Video observations	0.80	0.68	-

Table 6.6 *Pearson Product Moment Correlations Between Sleep Measures for Duration of Night Wakings*

	1	2	3
1. Parent-report sleep diary	-		
2. Self-report sleep diary	-	-	
3. Video observations	0.57	0.85	-

Table 6.7. *Pearson Product Moment Correlations Between Sleep Measures for Sleep Efficiency*

	1	2	3
1. Parent-report sleep diary	-		
2. Self-report sleep diary	0.87	-	
3. Video observations	0.82	0.82	-

Table 6.8. *Pearson Product Moment Correlations Between Sleep Measures for Total Sleep Time*

	1	2	3
1. Parent-report sleep diary	-		
2. Self-report sleep diary	0.72	-	
3. Video observations	0.56	0.21	-

Data Quality

As Eve's parents could not always detect covert wake behaviour (hence exclusion from IOA analysis) there were discrepancies between parent-report and video observed values across target behaviours. For this reason, video observations are reported to supplement parent-report. Eve completed self-report sleep diaries from night 23 to 54. These discrepancies notwithstanding, parent-report is the primary dependent variable reported, since there was more continuous data from this source than from the other sources.

Necessary treatment components

Eve met criterion for maintenance during the third phase of treatment (consisting of white noise, relaxation instruction, and stimulus control), therefore additional planned intervention phases involving reinforcement and extinction procedures were not implemented Overall, intervention lasted for 43 days.

Effect on CCs

Parent-reported CCs ranged from none to three per night in baseline, with most baseline nights having one CC (see Figure 6.1). There was significant variance in CC levels across parent-report, self-report, and video (parent-report range, 0 - 3; self-report range, 0 - 7; video range, 0 - 5). There was some reduction in CCs in the first treatment phase, but this was not maintained in the second phase. Only with the full combined treatment (Phase 3) was there consistent reduction in CCs to zero (or one per night at worst), and this was maintained at follow-up. The reduction in CCs achieved was clinically substantive, and stable (Phase 3 PBM $_{\text{parent-report}}$ = 91%, PBM $_{\text{video}}$ = 100%; follow-up PBM $_{\text{parent-report}}$ = 100%, PBM $_{\text{video}}$ = 100%).

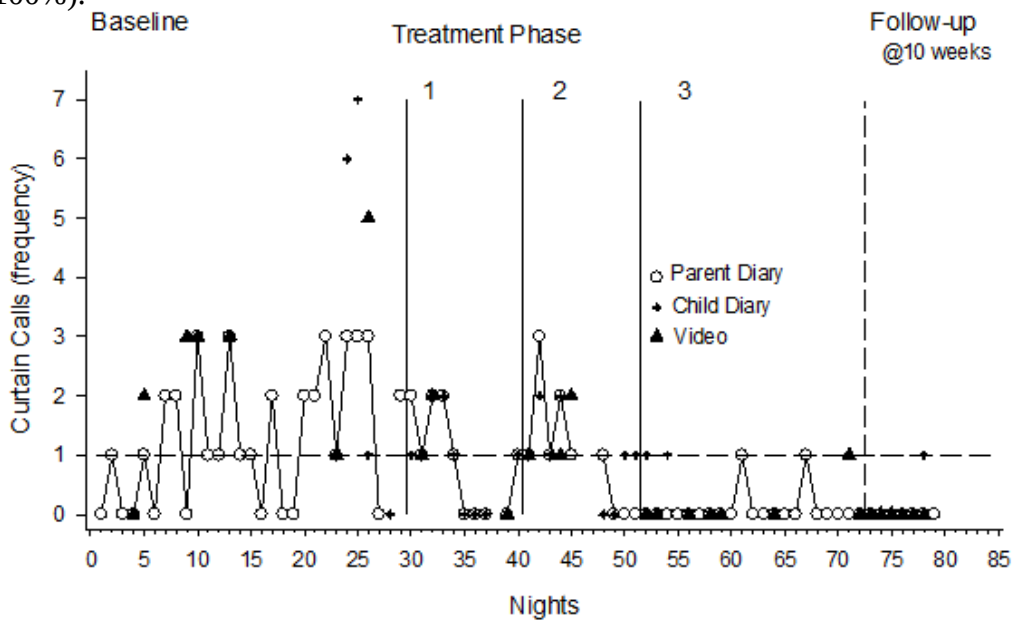

Figure 6.1. Frequency of Eve's curtain calls across baseline, intervention, and follow-up phases. Treatment Phase 1 = white noise; Phase 2 = white noise and relaxation instruction; Phase 3 = white noise, relaxation instruction, and stimulus control. The dashed horizontal line is the cut-off for clinical levels of curtain calls (≥1) engaged in by school-aged children.

Effect on SOL

Eve consistently experienced clinical levels (> 45 min) of SOL during baseline (parent-report median = 67.5 min, self-report median = 70 min, video median = 55 min), with a substantial number of nights having clinically severe levels (> 90 min, see Figure 6.2). There was little evidence of change in SOL during treatment Phase 1 or 2; only in Phase 3 was a clinically substantive reduction in SOL observed (PBM $_{\text{parent-report}}$ =100%, PBM $_{\text{self-report}}$ = 100%, PBM $_{\text{video}}$ = 100%). The reduction in median SOL from baseline to the final phase of treatment was -53 min (parent-report), -25 min (self-report), and -37 min (video). SOL reduction was maintained at follow-up (PBM $_{\text{parent-report}}$ =100%, PBM $_{\text{self-report}}$ = 100%, PBM $_{\text{video}}$ = 100%) as Eve fell asleep within 15 min each night.

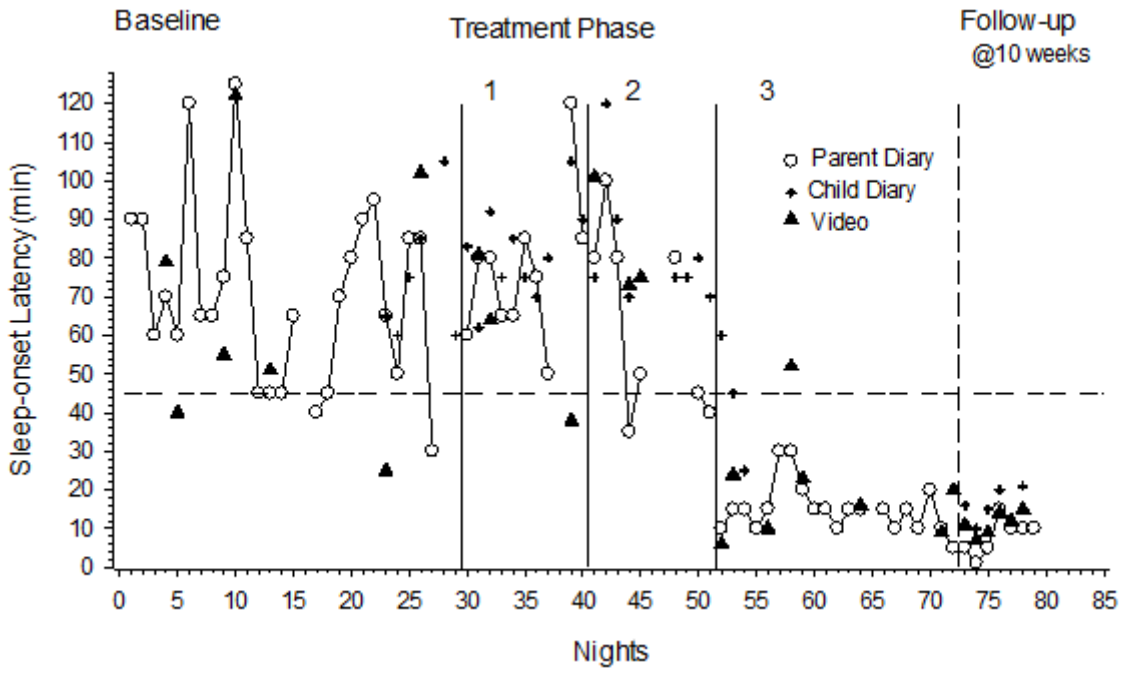

Figure 6.2. Eve's sleep onset latency across baseline, intervention, and follow-up phases. Treatment Phase 1 = white noise; Phase 2 = white noise and relaxation instruction; Phase 3 = white noise, relaxation instruction, and stimulus control. The dashed horizontal line is the

clinical cut-off for sleep onset delay (> 45 min) indicative of poor sleep in school-aged children.

Effect on NWs

Parent-report and self-report indicated the duration of Eve's NWs were not of clinical concern during baseline or at any point thereafter, but video recordings showed Eve woke regularly during baseline for extended time periods (range, 77 - 243 min; see Figure 6.3). Video data shows there was an immediate, clinically significant reduction in the duration of NWs only during Phase 3 (PBM $_{video}$ = 100%). The median video duration reduced from 104 min in baseline to 39 min in the final phase, however, the length of WASO was still of clinical concern on occasional nights during this phase. Video data demonstrated there was a further reduction in the duration of NWs during follow-up (video median = 16 min) and length was not of clinical concern on any night.

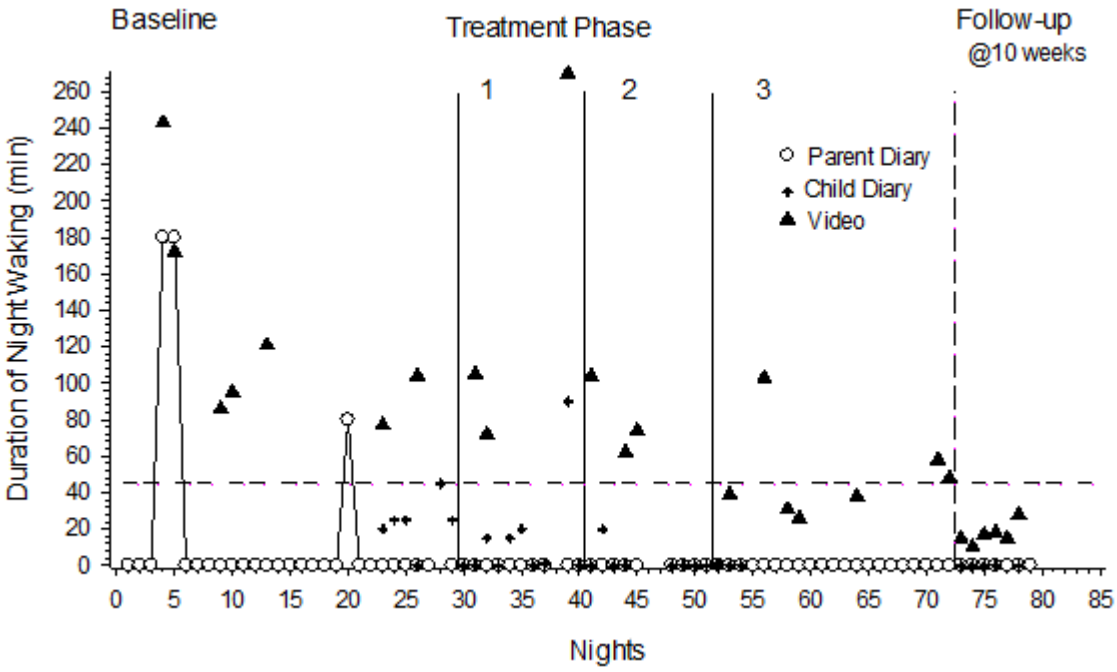

Figure 6.3. Duration of Eve's night wakings across baseline, intervention, and follow-up phases. Treatment Phase 1 = white noise; Phase 2 = white noise and relaxation instruction; Phase 3 = white noise, relaxation instruction, and stimulus control. The dashed horizontal line is the clinical cut-off for duration of night wakings (> 40 min) indicative of poor sleep quality in school-aged children.

Effect on Total Sleep Time and SE

Video data showed Eve's total sleep duration (video median = 444 min) was more problematic than parent-report or self-report indicated (parent-report median = 572.5 min, self-report median = 563 min), with a number of nights in baseline within the clinical severity range (< 420 min; see Figure 6.4). Nevertheless, there was still an improvement in parent-reported sleep duration within each treatment phase (PEM $_{\text{parent-report}}$ Phase 1 = 90%, Phase 2 = 86%, Phase 3 = 91%). Video data revealed both Phases 2 and 3 had large treatment effects (PEM $_{\text{video}}$ Phase 2 and 3 = 100%), which were clinically substantive, with median sleep duration increasing from 444 min in baseline to 525 min in the final phase. In addition, video data revealed Eve reached optimal sleep duration (540 to 660 min/ 9 to 11 hours) on occasion during treatment. Improvement in total sleep duration was maintained at follow-up (video median = 564 min) and Eve achieved optimal duration of sleep each night.

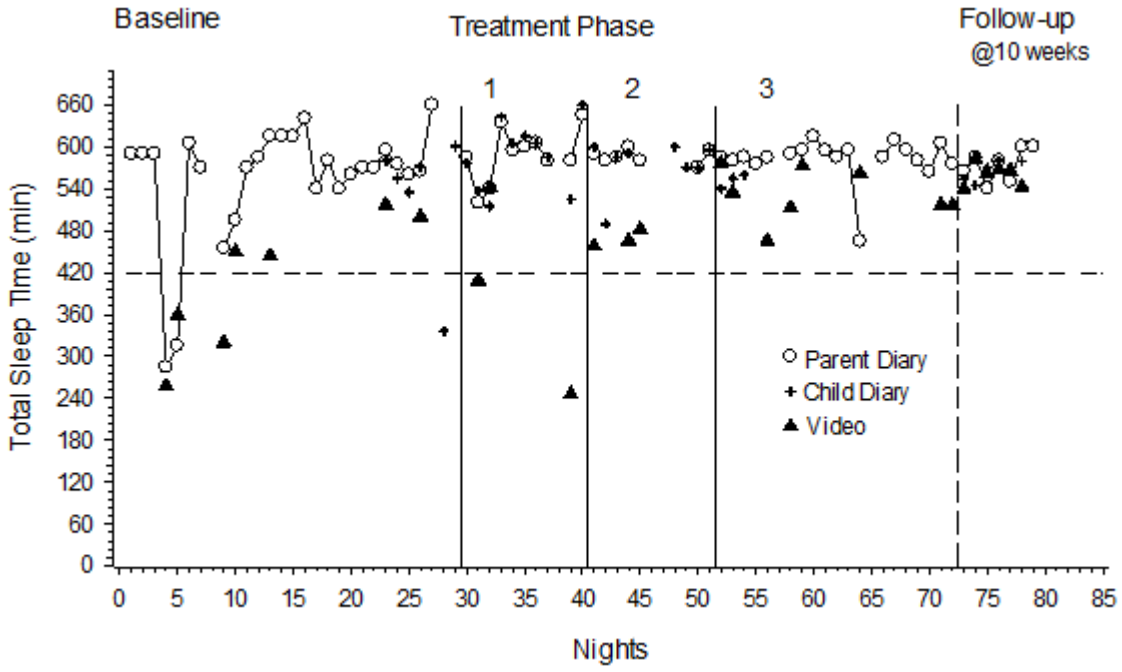

Figure 6.4. Eve's total sleep time across baseline, intervention, and follow-up phases. Treatment Phase 1 = white noise; Phase 2 = white noise and relaxation instruction; Phase 3 = white noise, relaxation instruction, and stimulus control. The dashed horizontal line is the clinical cut-off for poor sleep duration (420 min) in school-aged children.

Video data showed Eve had poor SE during baseline (video median = 67%), although parent- and self-report suggested it was less problematic (parent-report and self-report

median = 87%; see Figure 6.5). Both parent-report and video data indicated Eve's SE was highly variable (parent-report range, 47 - 96%; video range, 41 - 84%) during baseline and fell in the clinical range (< 75%) at times. Treatment Phase 1 had no effect on SE, but Phase 2 had a moderate to large effect (PEM $_{\text{parent-report}}$ = 86%, PEM $_{\text{video}}$ = 100%). There was further clinically substantive improvement in Phase 3 (PEM $_{\text{parent-report}}$ = 95%, PEM $_{\text{video}}$ = 100%) which was maintained at follow-up (PEM $_{\text{video}}$ = 100%).

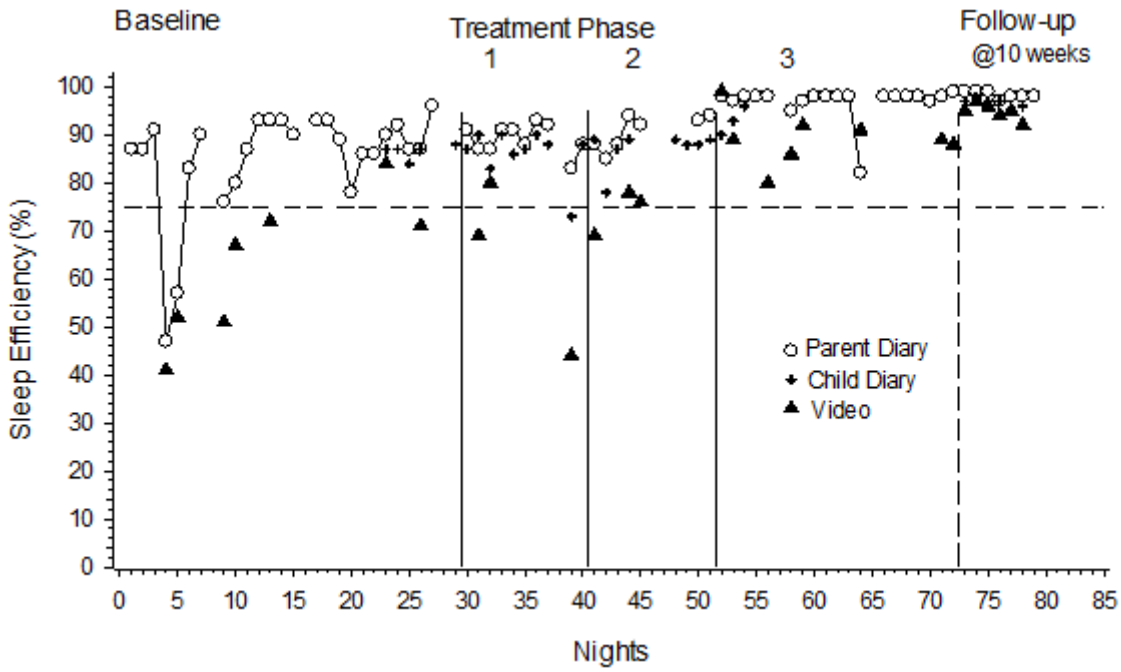

Figure 6.5. Eve's sleep efficiency across baseline, intervention, and follow-up phases. Treatment Phase 1 = white noise; Phase 2 = white noise and relaxation instruction; Phase 3 = white noise, relaxation instruction, and stimulus control. The dashed horizontal line is the clinical cut-off for poor sleep efficiency (< 75%) in school-aged children.

Overall Sleep Quality, CSHQ, and SSR

Parent report, self-report, and video observation showed Eve met criteria for poor sleep quality (as evidenced by reported clinical levels of any sleep variable) on only 25% of nights during Phase 3 and 0% of nights at follow-up, compared with 100% of baseline nights. The total CSHQ score reduced from pre- to post-treatment, however Eve's score remained within the clinical range (see Table 6.9). Parent ratings yielded large improvements in the Sleep Onset Delay and Sleep Duration domain scores, though there was little change in other

subscale scores. Eve's ratings yielded an improvement in the SSR total score and small improvements in the Sleep Onset Delay, Night Wakings, and Daytime Sleepiness subscale scores (see Table 6.10). There was no improvement reported in other subscale scores.

Table 6.9. *Children's Sleep Habits Questionnaire (CSHQ) Pre-and Post-treatment Scores*

CSHQ scales	Pre	Post
Bedtime Resistance	7	6
Sleep Onset Delay	3	1
Sleep Duration	7	4
Sleep Anxiety	7	6
Night Wakings	3	4
Parasomnias	7	8
Sleep-Disordered Breathing	3	3
Daytime Sleepiness	18	17
Total Score	51	47

Table 6.10. *Sleep Self Report (SSR) Pre- and Post-treatment Scores*

SSR Scales	Pre	Post
Bedtime Resistance	4	4
Sleep Onset Delay	2	1
Sleep Duration	2	2
Sleep Anxiety	3	3
Night Wakings	4	3
Daytime Sleepiness	3	2
Total Score	41	36

Treatment Fidelity and Child Social Validity

Treatment fidelity was high but tended to reduce over time. Mean treatment fidelity during Phase 1 was 100%, Phase 2 was 93% (67-100%), and Phase 3 was 70% (range, 60-100%). Mean treatment fidelity during follow-up was 80%. During the post-treatment interview Eve commented on the effectiveness of treatment, noting she could now fall asleep within 30 minutes at bedtime, within 20 minutes during NWs, and she no longer calls out or leaves her bedroom after bedtime. Eve considered delayed bedtime to be the most helpful and favoured treatment component. She enjoyed engaging in PMR as "it felt nice" but did not find the relaxation resource book helpful. She also disliked not using her e-reader in bed as this had long been part of her bedtime routine. According to the YPTE, Eve considered treatment to be moderately acceptable. YPTE scores have a possible range of 6 to 30, with higher scores indicating higher treatment acceptability. Eve's ratings yielded a total score of 18 (see Table 6.11).

Table 6.11. *Young Person Treatment Evaluation Scores*

Scale	Eve	Maximum Score
Effectiveness	6	10
Enjoyableness	3	5
Time	3	5
Fairness	3	5
Overall	3	5
Total	18	30

Parent Social Validity

Eve's parents noted the intervention strategies were easy to implement. Eve's mother felt that white noise and stimulus control were critical to intervention success. She reported providing Eve a sense of control over which sound to choose contributed to her acceptance of the white noise. Eve's mother thought stimulus control increased Eve's sleep pressure and reduced sleep-interfering behaviour, as she could not easily access her e-reader during NWs. The importance of actively including Eve within the therapeutic process was emphasised. Eve's mother noted the communication methods (e.g., letters) were effective in engaging Eve

and the incorporation of her interests helped Eve feel “positive” and “excited” about the study. Eve’s mother said “not treating her like a subject” by involving her (e.g., explaining treatment rationales) reduced Eve’s anxiety regarding the process. She also noted the improvement in Eve’s sleep had “affected the whole household”, all family members felt less stressed and irritable, and parent-child interactions were calmer. Additionally, Eve’s mother felt the intervention had a positive impact on Eve, who she described as feeling “happier” and “less anxious”. She also reported Eve’s focus and task completion during the morning routine had improved.

TARF-R scores have a possible range of 17 to 119, with higher scores indicating higher acceptability. Eve’s parents’ ratings both yielded a score of 100 (see Table 6.12). Overall, Eve’s parents indicated the intervention package as a whole was effective, reasonable, and low-cost. Although, was moderately time-consuming and disruptive to their regular routine.

Table 6.12. *Treatment Acceptability Rating Form-Revised Scores*

Scale	Mother	Father	Maximum Score
Effectiveness	20	19	21
Reasonableness	19	19	21
Willingness	18	20	21
Cost	12	14	14
Negative side-effect	17	17	21
Disruption/time	14	11	21
Problem severity*	11	9	14
Understanding of treatment*	6	7	7
Total acceptability	100	100	119

Note. *Not included in total acceptability score

Discussion

In this case study, FBA-informed treatment components were implemented sequentially to address sleep disturbance experienced by a 9-year-old girl with ASD and

selective mutism in a minimally sufficient and least restrictive manner. White noise alone had no effect on target sleep variables. White noise and relaxation instruction produced a statistically significant reduction in CCs, an increase in SE, as well as a clinically substantive improvement in total sleep duration also. White noise, relaxation instruction, and stimulus control produced statistically significant and clinically substantive improvements across all sleep variables. These improvements were maintained at 10-week follow-up. Eve and her parents considered the overall treatment package to be effective, reasonable, and affordable. Their preferred treatment components were white noise and implementation of a faded bedtime.

There is limited evidence for the treatment of sleep disturbance in children with ASD using white noise alone (McLay & France, 2016). Further, while relaxation instruction is commonly incorporated in cognitive behavioural anxiety treatment (Ho et al., 2015) and has been included in multicomponent sleep interventions for young people with ASD (Loring et al., 2016; McCrae et al., 2019; van Deurs et al., 2019), the efficacy of this technique alone has not been established. Application of individual treatment components alone are unlikely to be able to address the range of antecedent and consequence variables underlying sleep problems. The current study attempted to implement as few and least restrictive components as possible to effectively reduce sleep disturbance. White noise functioned to mask external noises purported to maintain anxiety (e.g., parental conversations) and that were disruptive to sleep (e.g., sibling crying). Relaxation instruction was intended to reduce hyperarousal, lessen the reinforcing value of parent-interaction post-bedtime, and facilitate sleep-conducive behaviour. Stimulus control functioned to ensure sufficient homeostatic sleep pressure, strengthen appropriate discriminative stimuli for sleep, and increase motivating operations for sleep. While white noise and relaxation instruction had some effect, the combination of white noise, relaxation instruction, and stimulus control were necessary to address the function of Eve's sleep-interfering behaviour.

Treatment components were ordered in accordance with behaviour function as well as from least to most restrictive and time intensive. Therefore, although lack of physiological sleep pressure was implicated in each of Eve's sleep problems, stimulus control was not introduced until Phase 3. This was because it is more time intensive than white noise or relaxation instruction and can be considered somewhat aversive in that it involves increased parent supervision in the evening (reducing parent alone-time) and meant Eve was not able to

use her e-reader in her preferred location. Consequence-based interventions, including the most restrictive and time-consuming fifth phase (white noise, relaxation instruction, stimulus control, reinforcement, and unmodified extinction), were not required.

This case study shows treatment components can be implemented in a sequential manner to ensure families are not required to engage in numerous unnecessary and restrictive strategies. For example, extinction of parent attention (i.e., modifying parental responses to CCs) was not required, although there was a strong attentional component to Eve's sleep disturbance. However, implementation of a multicomponent treatment from the outset may have resulted in faster progress.

In this case, implementation of fewer and less restrictive procedures did not necessarily improve treatment acceptability, compared with TARF-R ratings for comprehensive behavioural sleep interventions involving more restrictive practices (e.g., unmodified or modified extinction; McLay et al., 2017; McLay, France, Knight et al., 2019). Eve's parents still considered treatment to be relatively time-consuming and somewhat disruptive. Eve was resistant to using her bed for sleep only and disliked using her e-reader in an alternative setting. It is common for people on the autism spectrum to become distressed in response to changes in their typical routine. Behavioural sleep interventions by their very nature consist of changes to the child's typical sleep routine and environment. Consequently, less restrictive approaches may still be aversive for family members, particularly in the face of child resistance.

Intervention agents are less likely to comply with treatment procedures as response effort increases (Friman & Poling, 1995). Accordingly, in the current study treatment fidelity reduced during the final treatment phase, which consisted of the most components. Eve and her parents stopped completing relaxation strategies prior to bed and at times Eve was bid goodnight before the agreed faded bedtime. Eve disliked completing relaxation exercises and her parents thought it did not reduce purported hyperarousal. Additionally, the final phase coincided with parental illness. Seemingly, Eve and her parents minimised response effort by only implementing components they preferred and considered to be most effective. Although, this phase had a significant treatment effect, video observations showed SOL and duration of NWs still fell within the clinical range on occasion. The reduction in SOL and duration of NWs may have been larger had the treatment plan been followed with more integrity, as was observed during follow-up. This example highlights the importance of composing a treatment

plan which includes the fewest, preferred components necessary to produce behaviour change, which in turn may enhance treatment fidelity and facilitate maintenance.

Parent-report sleep diary data and video observation indicated there was significant and clinically substantive change in Eve's sleep. This was not reflected in parent- or self-report questionnaire results. Although CSHQ and SSR scores improved over time, the magnitude of difference was relatively small. Further, Eve's total CSHQ score remained in the clinical range post-treatment. Given the variable number of items within each CSHQ subscale (e.g., Sleep Onset = 1 item, Daytime Sleepiness = 8 items), Johnson et al. (2016) raise concern that the CSHQ may not adequately or reliably measure symptoms of sleep disturbance in ASD. Further, endorsement of specific CSHQ items, such as the child "wets the bed at night", or "is restless and moves a lot during sleep" may be related to the presence of a neurodevelopmental disorder, as opposed to indicative of sleep disturbance (Johnson, Katz et al., 2018). The CSHQ in its current form (validated on typically developing children) may not be the most effective measure of common sleep concerns for young people with ASD (Johnson et al., 2016). Several studies illustrate revised versions of the CSHQ with four- (Katz et al., 2018) or five-factor models (Johnson et al., 2016; Zaidman-Zait et al., 2020) compared with the original eight-factor structure, may be more appropriate. The newly proposed factors in these studies capture sleep disturbances common among young people with ASD, such as Bedtime Routine problems, Insufficient Sleep and Sleep-onset, as well as Co-sleeping and Sleep Anxiety (Johnson et al., 2016; Katz et al., 2018; Zaidman-Zait et al., 2020). The reliability and validity of these revised factor models require further examination.

Although Pearson product moment correlations indicated a relatively strong relationship between parent-report and video data, there were large discrepancies in the individual values reported each night (as can be observed in Figures 6.1 to 6.5). This is because Eve's parents were unable to detect the duration of target sleep variables (e.g., SOL, WASO) when she lay quietly in bed and did not seek them out. As children develop greater autonomy, parents may be less aware of covert sleep disturbances; cognitive and communicative abilities of young people with ASD may inhibit their ability to accurately self-monitor and subsequently report sleep behaviour to their parents. In this study, most self-report data did not correlate strongly with video data and IOA between these two measures was low. Eve may have had difficulty remembering numerous brief arousals in the morning. Previous research indicates children and adolescents have difficulty correctly evaluating

subjective sleep variables (e.g., duration of NWs; Bauer & Blunden, 2008). Identification of sleep/wake patterns may be even harder for children with ASD (Katz et al., 2018).

A number of limitations should be considered when interpreting the results of this study. Firstly, sequence effects may have impacted treatment outcomes (i.e., white noise, relaxation instruction, and stimulus control may have only been effective due to being preceded by white noise and white noise and relaxation instruction phases). Although results suggest stimulus control was the primary active component within the FBA-informed treatment package, it is not possible to conclude whether stimulus control would have been sufficient alone. Sequence effects were not able to be mitigated by employing reversal conditions between phases, given relaxation instruction resulted in skill improvement which could not be eliminated. Secondly, as follow-up data was collected only once at 10 weeks post-treatment the long-term maintenance of this intervention was not established. Thirdly, while a minimally sufficient approach was effective in eliminating sleep disturbance for the participant within this study, this approach may not be appropriate or effective for other children on the autism spectrum with diverse sleep presentations. Replication with larger samples whereby sequence effects are accounted for and long-term maintenance is evaluated, is necessary to demonstrate the efficacy of minimally sufficient FBA-informed sleep interventions. Future research could implement treatment components individually to evaluate the necessity and sufficiency of each component alone, before combining components into a treatment package. This is critical to identify active treatment components and ensure clinicians and families can implement the least restrictive and minimally sufficient interventions necessary to treat sleep problems, improving social validity and treatment maintenance.

Chapter 7: Psychometric Outcomes

Results presented in the preceding three studies illustrated that intervention was generally effective in reducing or eliminating sleep disturbance (the primary outcome) for all participants, according to sleep diaries, VSG, parent report, and participant report. In addition to these measures, a range of questionnaires were completed by participants and their parents pre- and post-treatment to assess secondary outcomes for participant and parent wellbeing. Copious research has demonstrated sleep disturbance is associated with detrimental effects on individual and family functioning, therefore, sleep interventions may benefit overall wellbeing. One of the aims of the current research is to assess the collateral impact of the selected interventions. This may assist health professionals in optimising psychological treatment for young people with ASD and their families. This chapter presents pre- and post-treatment sleep outcome data gathered from questionnaires as well as reported change in secondary outcomes.

Procedure

Participants and their parents were provided with paper versions of pre-treatment questionnaires once eligibility to participate in the overall study was established. Participants were instructed to complete questionnaires independently, with their caregivers help if desired, while parents were instructed to independently complete questionnaires pertaining to their own wellbeing, with the exception of one parent with eye-sight difficulties who was administered all questionnaires over the phone by the researcher. In two-parent households either caregiver completed parent-report questionnaires regarding their child. Questionnaires were posted back before baseline/treatment commenced. Immediately upon conclusion of treatment, the same battery of questionnaires were posted to participants and their parents and completed in the same way. To ensure rating consistency, the same caregiver who completed parent-report questionnaires for their child completed the forms at post-treatment. Questionnaires were returned to the researcher within 6 weeks of treatment finishing. Respondent failure to complete each questionnaire item, inconsistent contact, and limited psychometric measure availability led to some missing data.

Sleep Outcome Measures

CSHQ. The CSHQ was completed by 7/12 parents pre- and post-treatment to evaluate change in parent-reported sleep disturbance (see Data Quality section, p.172, for an

explanation of respondent numbers). The CSHQ is a parent-report questionnaire consisting of 45 items relating to children's sleep patterns (Owens, Spirito, & McGuinn, 2000). Parents or caregivers are required to indicate the frequency of specific behaviours in a typical week using a 3-point Likert scale, 3 = "usually" (5 to 7 times a week), 2 = "sometimes" (2 to 4 times a week) and 1 = "rarely" (0 to 1 time a week, Owens, Spirito, & McGuinn, 2000). A higher score is indicative of poorer sleep and scores ≥ 41 are indicative of clinically significant sleep disturbance (Owens, Spirito, & McGuinn, 2000). The CSHQ consists of eight subscales which assess common sleep problems experienced by young children: Bedtime Resistance, Sleep Onset Delay, Sleep Duration, Sleep Anxiety, Night Wakings, Parasomnias, Sleep Disordered Breathing, and Daytime Sleepiness (Owens, Spirito, & McGuinn, 2000). The CSHQ has satisfactory internal consistency (α = 0.68 to 0.78) and test-retest reliability (r = 0.62 to 0.79) for both community and clinical populations respectively (Owens, Spirito, & McGuinn, 2000). Psychometric properties used to assess the reliability and validity of the measure, as well as calculate the Standard Error of the measure and hence the RCI, were derived from a community sample of 494 children (4 to 11 years) in Rhode Island, U.S.A (Owens, Spirito, & McGuinn, 2000). Peter, John, and Isaac's parents were not required to complete the CSHQ as these participants were not within the measure's validated age range.

The SSR. The SSR was completed by four participants to evaluate change in child-reported sleep. The SSR is a 26-item self-report questionnaire for children 7 to 12 years of age which corresponds with the CSHQ subscales (Owens, Spirito, McGuinn & Nobile, 2000). Within the current study participants reported on the frequency of specific sleep behaviours engaged in over the past week using a 3-point Likert scale, 3 = "usually" (5 to 7 times a week), 2 = "sometimes" (2 to 4 times a week) and 1 = "rarely" (0 to 1 time a week; Owens, Spirito, McGuinn & Nobile, 2000). SSR scores have a possible range of 23 to 69 with higher scores indicative of worse sleep (no clinical cut-off has been established). The SSR has been shown to have adequate discriminative validity and test-retest reliability (r = 0.76 - 0.88) among both clinical and non-clinical American, Spanish, and Dutch populations (Orgilés et al., 2013; Owens, Spirito, McGuinn & Nobile, 2000; Steur et al., 2019). The SSR was not completed by pilot participants, or by young people outside of the validated age range.

The Children's Sleep Comic (CSC; Schwerdtle, Kanis, Kahl, Kübler & Schlarb, 2012). The CSC was completed by four participants to gather qualitative information about

their sleep (e.g., pre-bedtime activities), as well as additional quantitative data. The CSC is a 20-item illustrated questionnaire that is presented within a comic book format. It assesses the following aspects: sleep hygiene, sleep quality, nighttime fears, dreaming, morning waking, nighttime sweating, nighttime bruxism, daytime napping, daytime activities, and somatic complaints (Schwerdtle et al., 2012). Children are required to tick square boxes underneath each pictured item to indicate whether the statement applies to them (see Figure 7.1). Response options are classified as "positively supporting sleep", "negatively affecting sleep", or "neutral". Any behaviours which are classified as "negatively affecting sleep" are scored a one. Scores range from 0 to 20, with higher scores indicative of worse sleep. Scores ≥ 9 are indicative of insomnia, however sensitivity (57%) and specificity (75%) are relatively low for this value (Schwerdtle et al., 2016). The CSC can be self-administered or administered to children 5 to 11 years of age. The CSC has been shown to have adequate construct validity and test-retest reliability (r = 0.83, Schwerdtle et al., 2012). The RCI for the CSC is 4. The CSC was not completed with pilot participants, or with young people outside of the validated age range.

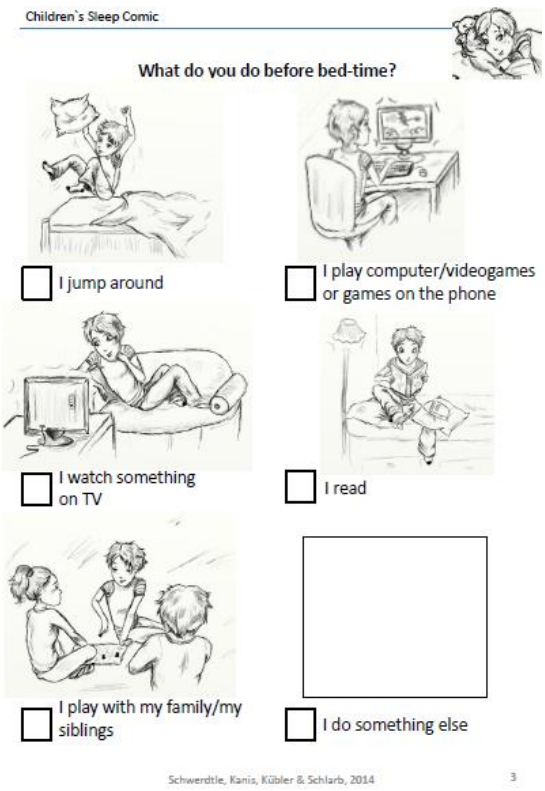

Figure 7.1. An example item from the Children's Sleep Comic. From "Children's Sleep Comic", by B. Schwerdtle, J. Kanis, A. Kübler, & A. A. Schlarb, 2014, p. 3. n.p: n.p.

Adolescent Sleep Hygiene Scale (LeBourgeois, Giannotti, Cortesi, Wolfson, & Harsh, 2005). The Adolescent Sleep Hygiene Scale (ASHS) was used to evaluate change in adolescent-reported sleep disturbance. It was completed by two adolescent participants pre- and post-treatment. The ASHS is a 32-item self-report questionnaire for 12- to 18-year-olds used to assess the frequency of sleep-interfering behaviour (e.g., "I fall asleep while watching television"). Respondents report the frequency of their own sleep-related behaviours over the past month on a 6-point Likert scale (1 = "always," 2 = "frequently-if not always," 3 = "quite often," 4 = "sometimes," 5 = "once in a while," and 6 = "never"). The mean of each subscale is calculated, and the aggregate mean of all subscales provides the total score. Scores range from 1 to 6, with higher scores indicative of better sleep hygiene. Five items were adapted to suit a modern NZ context and limit potential literal interpretations[4]. Reliability is low for some subscales but is high for the total score ($\alpha = 0.80$). Consequently, only the ASHS total score was analysed pre- and post-treatment. The RCI for the ASHS total score is 0.5. Psychometric properties were derived from a sample of 572 adolescents (12 to 17 years) living in Mississippi (LeBourgeois et al., 2005).

Adolescent Sleep Wake Scale Revised (Essner, Noel, Myrvik, & Palermo, 2015). The Adolescent Sleep Wake Scale Revised (ASWS-R) was used to assess change in adolescent-reported sleep patterns. It was completed by two adolescent participants pre- and post-treatment. The ASWS-R is a short version of the original 28-item ASWS and was chosen to reduce the time burden of completing questionnaires. The ASWS-R consists of 10 items which assess the following domains: Falling Asleep and Reinitiating Sleep, Returning to Wakefulness, and Going to Bed. Similar to the ASHS, adolescents report the frequency of their own sleep-related behaviour over the past month on a 6-point Likert scale (1 = "always," 2 = "frequently-if not always," 3 = "quite often," 4 = "sometimes," 5 = "once in a

[4] The changes were as follows (altered wording is italicised): "I have drinks with caffeine (for example: cola, root beer, iced tea)" became "I have drinks with caffeine (for example: *coke, energy drinks*, iced tea, *coffee*)"; "I fall asleep in a brightly lit room (for example: the overhead light is on) became "I fall asleep in a brightly lit room (for example: the *main* light is on)"; "I do things that make me feel very awake (for example: talking on the telephone, watching television, playing video games, doing homework)" became "I do things that make me feel very awake (for example: playing video games, watching television, *using my phone*); "I do things in my bed that keep me awake (for example: watching television, reading)" became "I do things in my bed *or bedroom* that keep me awake (for example: watching television, reading)"; I use my bed for things other than sleep (for example: talking on the telephone, watching television, playing video games, doing homework)"became "I use my bed for things other than sleep (for example: *watching video clips or shows*, playing video games, doing homework)"; finally "I check my clock several times during the night" became "I check the *time* several times during the night".

while," and 6 = "never"). The mean of each subscale is calculated, and the aggregate mean of all subscales is the total score. Scores range from 1 to 6, with higher scores indicative of better sleep. The ASWS-R has been validated for use with ethnically and economically diverse adolescents. Internal validity is high for the total score (α = 0.72 – 0.81) and two of the sleep domains (α = 0.74 – 0.89), however it is low for the Going to Bed subscale (α = 0.48 – 0.74, Essner et al., 2015; Sufrinko et al., 2015). Psychometric properties were derived from a sample of 152 Caucasian adolescents (12 to 18 years) from an economically disadvantaged, rural community in the U.S.A (Sufrinko et al., 2015). The RCI for the ASWS-R is 1.

Secondary Outcome Measures - Young Person

GARS-3. The GARS-3 was completed by 11/12 parents pre- and post-intervention. This instrument was used to evaluate the impact of sleep interventions on behaviours symptomatic of ASD, such as stereotypy. The GARS-3 is a 58-item informant rating scale designed to assess the likelihood of autism and symptom severity in people aged 3 to 22 years (Gilliam, 2014). Ratings on a 4-point Likert scale (0 = "not at all like the individual, 1 = "not much like the individual", 2 = somewhat like the individual, and 3 = "very much like the individual") across six subscales (Restricted/Repetitive Behaviors, Social Interaction, Social Communication, Emotional Responses, Cognitive Style and Maladaptive Speech) are summed, yielding an overall ASD Index score, with higher scores indicative of more severe symptoms. In accordance with severity level descriptors within the DSM-5 the ASD Index score can be classified as Level 1 (Minimal Support Required), Level 2 (Requiring Substantial Support), or Level 3 (Requiring Very Substantial Support, Gilliam, 2014). Probability estimates are classified as Unlikely, Probable, or Very Likely. No severity level classification is necessary for individuals whose ASD Index score falls within the Unlikely probability range. The GARS-3 was standardised on an American sample of 1,859 people (3 to 22 years) with ASD and has been shown to have high reliability (internal consistency [α = 0.79 – 0.94], test-retest reliability [r = 0.76 – 0.90], and interrater reliability [r = 0.71 – 0.85]) and validity (Gilliam, 2014). The RCI for the GARS-3 ASD Index is 11.

Child Behavior Checklist for Ages 6–18 (CBCL; Achenbach, 2001). The CBCL was completed by 11/12 parents to evaluate participant's externalising and internalising behaviour, as well as competence within social, academic, and extracurricular activities pre- and post-treatment. The CBCL contains 113 items pertaining to problem behaviour

experienced by young people aged 6 to 18 years. Parent ratings on a 3-point Likert scale (0 = "not true", 1 = "somewhat or sometimes true", and 2 = very true or often true") across 9 subscales (Anxious/Depressed, Withdrawn/Depressed, Somatic Complaints, Rule-breaking Behaviour, Aggressive Behaviour, Social Problems, Thought Problems, Attention Problems, and Other Problems) are summed to provide composite scores for Internalising, Externalising and Total Problem behaviour scales (Achenbach & Rescorla, 2001). Higher scores on these scales are indicative of worse problem behaviour. The CBCL also contains 16 items which assess parent perception of their child's participation and performance in extracurricular activities, social activities, and school, yielding a composite score on the Competence scale (Achenbach & Rescorla, 2001). Lower scores on this scale are indicative of less competent behaviour. Overall, the CBCL has strong construct validity, adequate internal consistency (α = 0.63 to 0.79) and high test-retest reliability (r = 0.90) for problem behaviour and competence scale scores. Normative data for the CBCL was derived from a nonclinical sample of 1,753 ethnically and socioeconomically diverse young people from the U.S.A (Achenbach & Rescorla, 2001).

MASC-2. The MASC-2 Self-Report (MASC-2 SR) and Parent-Report (MASC-2 PR) was completed by six participants and five parents respectively. The questionnaire was completed pre- and post-treatment to evaluate change in self-reported and parent-reported anxiety levels. The MASC-2 is a 50-item multi-informant questionnaire designed to assess anxiety symptoms experienced by people aged 8 to 19 years. Respondents rate the frequency of anxiety symptoms experienced on a 4-point Likert scale (0 = "never", 1 = "rarely", 2 = "sometimes", and 3 = "often"). Ratings are summed to provide composite scores on six Anxiety Scales (Separation Anxiety/ Phobias, General Anxiety Index, Social Anxiety Total, Obsessions and Compulsions, Physical Symptoms Total, and Harm Avoidance) as well as a Total Score. The Social Anxiety Total Scale and Physical Symptoms Total Scale are comprised of Humiliation/Rejection and Performance Fears, and Panic and Tense/Restless subscales respectively. Higher scores on each scale are indicative of increased anxiety symptoms. Both the MASC-2 SR and PR have high internal consistency, test-retest reliability (r = 0.80 – 0.94), and discriminative validity (March, 2012). Psychometric properties of the MASC-2 SR and PR were obtained from normative samples of 1,800 young people and 1,600 parents' representative of U.S.A and Canadian populations (March, 2012).

Secondary Outcome Measures - Parents

Pittsburgh Sleep Quality Index (PSQI; Buysse, Reynolds, Monk, Berman, & Kupfer, 1989). The PSQI was completed by 19/21 parents to assess their own sleep quality pre- and post-treatment. This was intended to reveal whether changes in participant sleep were related to parental sleep. The PSQI is a 19-item adult self-report measure of sleep quality and disturbance (Buysse et al., 1989). Respondents are required to answer questions about their sleep over the past month (Buysse et al., 1989). Scores on seven components (sleep quality, sleep latency, sleep duration, habitual sleep efficiency, sleep disturbances, medication use, and daytime dysfunction) yield a global PSQI score (Buysse et al., 1989). Higher scores are indicative of poorer sleep, and scores > 5 distinguish poor from good quality sleepers (Buysse et al., 1989). Psychometric properties of the PSQI were derived from a sample of 138 adults (including healthy controls, individuals with major depressive disorder in inpatient and outpatient settings, and individuals referred to a sleep clinic for insomnia or excessive daytime sleepiness). The RCI for the PSQI is 6. Overall, the PSQI has been shown to have acceptable reliability and validity, however convergent validity (association between PSQI scores and PSG data) was low (Buysse et al., 1989).

Depression Anxiety and Stress Scale- 21 (DASS-21; Henry & Crawford, 2005). The DASS-21 was completed by 19/21 parents pre- and post-treatment to examine change in parent wellbeing over the course of intervention. The DASS-21, a short version of the 42-item Depression Anxiety and Stress Scale (DASS, Lovibond & Lovibond, 1993), is a 21-item self-report questionnaire designed to assess adult psychological distress in relation to the dimensions of depression, anxiety, and stress. Respondents are required to indicate the extent to which they have experienced such symptoms over the past week on a 4-point Likert scale (0 = “never”, 1 = “sometimes”, 2 = “often”, and 3 = “almost always”). Participant ratings yield scores on each dimension, as well as a Total Scale score. The DASS-21 is not a diagnostic tool; however, it illustrates the severity of symptoms felt by respondents, with higher scores indicative of increased severity. The DASS-21 has been demonstrated to have high internal consistency ($\alpha = 0.82 – 0.93$) and convergent and discriminant validity (Henry & Crawford, 2005). Psychometric properties were derived from a non-clinical community sample of 1,794 adults (18 to 91 years) in the U.K (Henry & Crawford, 2005). The RCI for each of the Depression, Anxiety, and Stress scales is 4 and the Total scale is 6.

Relationship Quality Index (RQI; Norton, 1983). Five mother-father dyads completed the RQI pre- and post-treatment to evaluate change in relationship quality over the course of intervention (it was not completed by caregivers residing in single-parent households). The RQI, also known as the Quality Marriage Index, is a 6-item self-report questionnaire relating to relationship quality and satisfaction (Norton, 1983). Five items are rated on a 7-point Likert scale (1 = "very strongly disagree", 2 = "very strongly disagree", 3 = "disagree", 4 = "neither agree nor disagree", 5 = "agree", 6 = "strongly agree", and 7 = "very strongly agree"). The final item, relating to global relationship satisfaction, is rated on a 10-point Likert scale ranging from "Unhappy" to "Perfectly Happy". Items are summed to provide a total RQI score. Scores range from a minimum of 6 to a maximum of 45, with higher scores indicative of greater overall relationship quality. Scores ≤ 29 are indicative of relationship distress. The RQI has adequate internal consistency ($\alpha = 0.68 – 0.85$) and strong convergent and discriminant validity (Norton, 1983). Psychometric properties were derived from a non-clinical sample of 430 couples from the Midwest region of U.S.A (Norton, 1983).

Data Analysis

Modified Brinley plots. In the current study, modified Brinley plots were used to evaluate change in psychometric scores from pre- to post-treatment (Blampied, 2017; Jacobson & Truax, 1991). A modified Brinley plot is a scatterplot which depicts the values of a single dependent variable at two time points and enables the evaluation of change at both the individual and group level (Blampied, 2017). This highlights the direction and degree of individual change, as well as replication of change across participants (Blampied, 2017). For any specific measure, pre-treatment (time 1; t_1) scores are plotted on the *x*-axis and post-treatment scores (time 2, t_2) are plotted on the $\boldsymbol{\gamma}$-axis. If there is no change in the value of the dependent variable from time 1 to time 2, the data points should lie on the 45° diagonal line (t_1 score = t_2 score). Data points will deviate from this 'line of no change', if pre- and post-treatment scores differ. The RCI is used to ascertain whether the degree of change was reliable (i.e., not due to measurement error) by specifying a minimum raw score change in dependent variable units. RCI boundaries are indicated by parallel lines above (+RCI) and below (-RCI) the 'line of no change' and reliable change has occurred when data points lie outside of these boundaries. In addition, clinical cut-off scores are displayed (when available) to illustrate clinically substantive change (i.e., scores changing from the clinical to non-clinical range). Figure 7.2 provides an example of a modified Brinley plot interpretation

using clinical cut-off points (as illustrated by a vertical line at t_1 and a horizontal line at t_2) which divide the plot into zones of change. Figure 7.3 further illustrates modified Brinley plot interpretation using clinical cut-off points and RCI boundaries (classification of the degree of change is illustrated by shading). Score difference over time is classified as an improvement or deterioration depending on the direction of therapeutic change. Figure 7.3 highlights when t_1 scores are close to the clinical cut-off participants can experience a clinical but non-reliable change in t_2 scores (this is indicated by the smallest triangle zones of change).

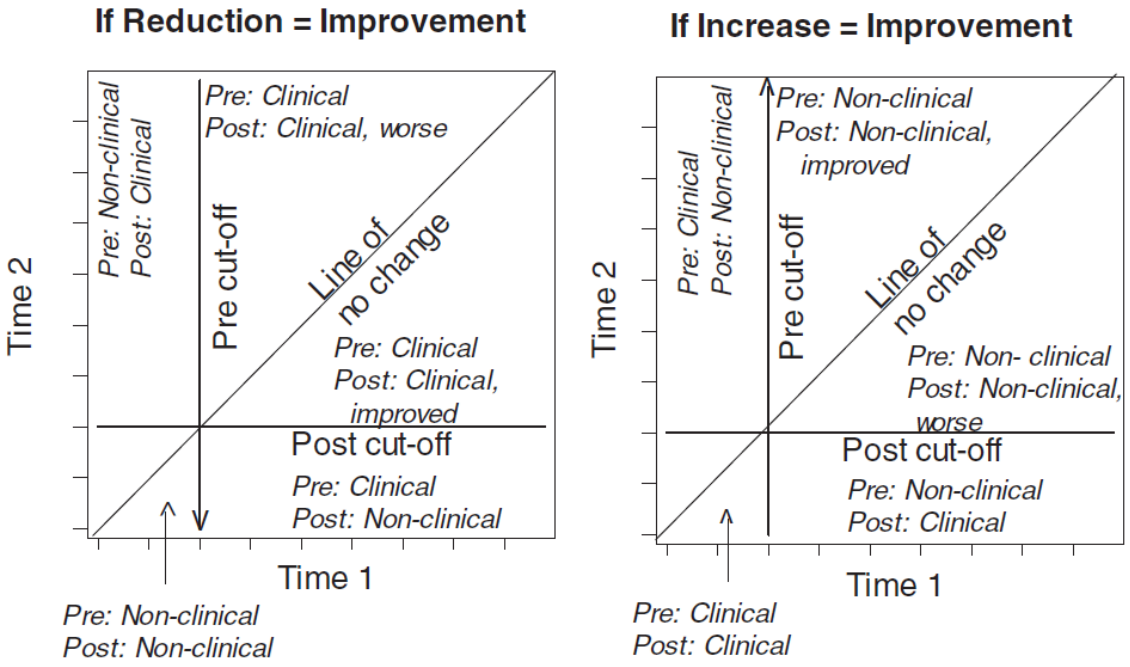

Figure 7.2. Examples of modified Brinley plots displaying clinical cut-off lines (vertical and horizontal lines), with zones of change created by the intersection of the cut-off lines and the 45° diagonal labelled to assist with interpreting the magnitude of individual change from Time 1 to Time 2. The arrow on the vertical clinical cut-off line indicates the direction of clinically desired change. From "Analyzing Therapeutic Change Using Modified Brinley Plots: History, Construction, and Interpretation", by N. M. Blampied, 2017, *Behavior Therapy, 48,* p. 120.

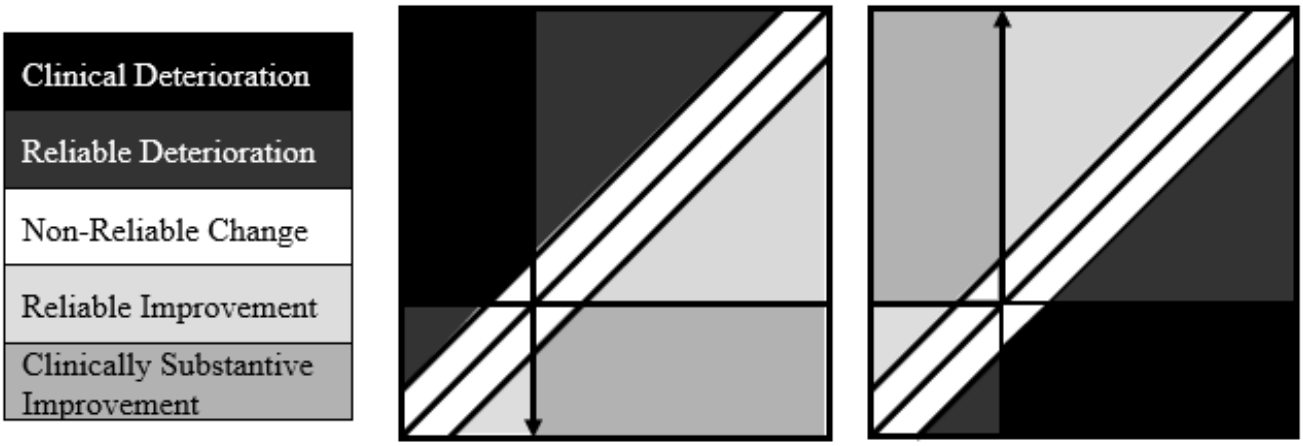

Figure 7.3. An example of modified Brinley plot interpretation using clinical cut-off points and RCI boundaries, with zones of change shaded. The arrow on the vertical clinical cut-off line indicates the direction of clinically desired change. Otherwise, interpretation is as for Figure 7.2

Effect size. Two effect size measures, Cohen's d_{av} and Common Language Effect Size (CLES; also known as the Probability of Superiority [PS]) were computed to further illustrate the practical significance of the results. Cohen's *d* (also known as the standardized mean difference effect size) describes a family of effect sizes commonly reported to illustrate the magnitude of difference between or within groups (Lakens, 2013). Given the current study involves repeated measures/paired data Cohen's d_{av} is the appropriate version of the effect size, calculated as shown in Equation 2 (where M_{diff} = the $Mean_{Pre}$ – $Mean_{Post}$ and the denominator is the mean of the respective standard deviations; Cumming, 2012).

$$\text{Cohen's } d_{av} = \frac{M\text{diff}}{\left(\frac{SD1+SD2}{2}\right)} \quad (2)$$

In accordance with Cohen's (1988) recommendations, effect sizes were interpreted as small (d = 0.20), medium (d = 0.50), and large (d = 0.80). The 95% confidence interval (CI) was calculated for Cohen's *d* to illustrate the precision of the effect size calculation and assess whether the difference between pre- and post-treatment scores was reliably different from 0 (Cumming, 2012). The CLES/PS is the likelihood (expressed as a percentage) that any randomly selected case has a post-treatment score that is clinically better than their pre-treatment score (Lakens, 2013; McGraw & Wong, 1992).

Reliable change. The degree of reliable change for each individual case is based on the raw difference score pre- and post-treatment and evaluated case-by-case. This was calculated for each participant and their parents when sufficient psychometric information,

namely Cronbach's alpha and the standard deviation of an appropriate reference group, was available in the literature. This data was used to compute the Standard Error of the difference scores (S_{Diff}; see Jacobson & Truax, 1991) and S_{Diff} was used in turn to standardise each individual's raw change score. Individual standardised change scores exceeding ±1.96 indicate reliable change from pre- to post-treatment; alternatively, $S_{Diff} \times 1.96$ gave the +/- RCI or raw change score needed for reliability to be judged. Pre- and post-treatment psychometric raw scores were used to assess reliable change as opposed to standard scores (e.g., T scores) as multiple participants changed norm groups (i.e., had a birthday) during the administration.

Results

The standardised change score for primary and secondary variables of interest are presented in Tables 7.2 to 7.13. Shading is used to facilitate interpretation of the degree of reliable and clinical change, data permitting (the key is illustrated in Figure 7.4). When insufficient psychometric data (e.g., Cronbach's alpha for normed population) was available to calculate reliable change, pre- and post-treatment psychometric raw scores were presented (Tables 7.3 and 7.13 [SSR and RQI Tables]). Modified Brinley plots were constructed when datasets for primary and secondary outcomes were sufficient (> 6 participants) to allow meaningful idiographic and nomothetic interpretations. Pre- and post-treatment scores are presented in modified Brinley plots for the CSHQ, GARS-3, DASS-21, PSQI, and RQI. Data is displayed in tables only for the SSR, CSC, ASHS, ASWS-R, MASC-2 SR, and MASC-2 PR. As the RCI and clinical cut-off parameters vary for the CBCL according to age and gender, and thus could not be plotted on a modified Brinley plot displaying each participants' data, reliable and clinical change are also presented in tables only.

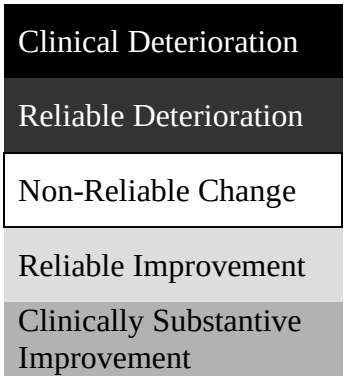

Figure 7.4. Reliable and Clinical Change Key

Data Quality

Reliable change could not be calculated for the SSR and RQI as information regarding psychometric properties (e.g., SD of a normed population) was not available within the literature. Insufficient information was available to calculate reliable change for the Sleep Onset Delay subscale of the CSHQ. The 95% CI for Cohen's d_{av} was not calculated for psychometric data sets with less than six pairs of participant data (SSR, CSC, ASHS, ASWS-R, MASC-2 SR and MASC-2 PR) as the precision could be compromised and render the data unreliable (Cumming & Finch, 2001).

Will and his parents did not complete post-treatment questionnaires and were therefore excluded from the analysis in this chapter because he experienced an improvement in sleep during assessment. Therefore, pre-treatment questionnaires did not provide an accurate baseline reference of primary or secondary outcomes. Blair and Peter participated in the study when the battery of pre- and post-treatment questionnaires were not as extensive, therefore they are not included in analyses of primary outcomes via questionnaires (e.g., CSHQ, SSR, CSC, ASHS, and ASWS-R). The MASC-2 SR and PR was not completed by pilot participants or their parents. In addition, Blair's parents did not complete the GARS-3, CBCL, or PSQI. Missing GARS-3 data prevented calculation of reliable change for this measure for Ben. Missing MASC-2 SR and PR data prevented reliable change calculations for Seth and Finn, including the Total Score. Missing data on the PSQI prevented calculation of reliable change for Seth and Ben's fathers and pre- and post-treatment RQI scores for Blair and Peter's parents could not be obtained due to missing data. Isaac completed the MASC-2 SR, however no parent-report forms were available at this time.

Primary Sleep Outcomes

Overview. Table 7.1 below provides an overview of parent- and participant-reported change in sleep according to CSHQ, SSR, CSC, ASHS, and ASWS-R. All participants (7/7) experienced a reduction in parent-reported sleep problems according to the CSHQ. All participants also indicated their sleep had improved according to the CSC (4/4), and SSR (3/3). Adolescent participants, John and Isaac, indicated improvement in their mean sleep hygiene score on the ASHS. Isaac's responses yielded an improvement in ASWS-R scores, whereas John's responses yielded a deterioration. Individual questionnaire results are presented in more detail below.

Table 7.1. *Pre and Post-treatment Sleep Questionnaire Scores*

	CSHQ		SSR		CSC		ASHS		ASWS-R	
	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post
Niko	52	44	-	-	-	-	-	-	-	-
Eric	58	38	-	-	-	-	-	-	-	-
Seth	60	58	-	-	-	-	-	-	-	-
Finn	47	45	43	28	10	5	-	-	-	-
Scott	64	39	38	25	10	1	-	-	-	-
Ben	47	41	-	-	8	5	-	-	-	-
Eve	51	47	41	36	10	7	-	-	-	-
John	-	-	-	-	-	-	4.14	4.40	4.40	4.10
Isaac	-	-	-	-	-	-	5.14	5.33	4.30	4.90

Note. CSHQ = Children's Sleep Habits Questionnaire; SSR = Sleep Self-Report; CSC = Children's Sleep Comic; ASHS = Adolescent Sleep Hygiene Scale; ASWS-R = Adolescent Sleep Wake Scale Revised.

CSHQ. As seen in Figure 7.5 all participants were in the CHSQ clinical range for sleep disturbance pre-treatment but, at post-treatment, all participants had reduced their CSHQ Total Score. Table 7.2 illustrates this improvement was reliable (RCI > 5) for four participants, and clinically substantive for two of the four participants. At post-treatment, five out of seven participants reported a reliable reduction in their Sleep Duration scale score but, (surprisingly), two out of the seven reported a reliable deterioration in their Sleep-Disordered Breathing score. Overall, the effect size was large for the CSHQ Total Score (d_{av} = -1.40) and reliably different from 0 (95% CI [-2.70, -0.11]). The likelihood that a randomly selected participant had an improved CSHQ score at post-treatment compared to pre-treatment was 85%.

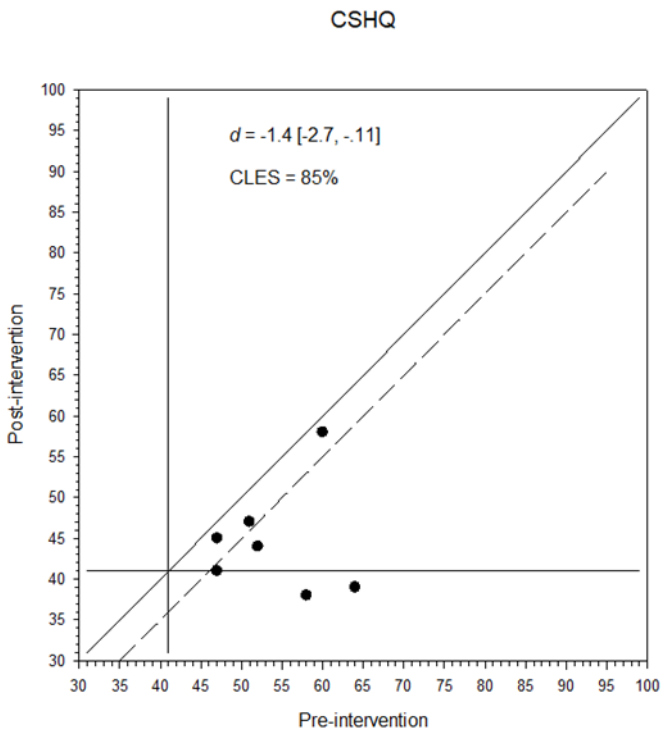

Figure 7.5. Modified Brinley plot showing change from pre- to post- intervention on the Children's Sleep Habits Questionnaire (CSHQ). *d* = Cohen's d_{av} effect size; CLES = common language effect size.

Table 7.2. *Children's Sleep Habits Questionnaire (CSHQ) Standardised Change Scores with Reliable and Clinical Change Shaded*

CSHQ Scales	Niko	Eric	Seth	Finn	Ben	Scott	Eve
Bedtime Resistance	0.00	-1.90	0.95	0.00	-0.95	-10.47	-0.95
Sleep Onset Delay	-	-	-	-	-	-	-
Sleep Duration	-5.17	-6.90	-3.44	-6.90	0.00	0.00	-5.17
Sleep Anxiety	-1.14	0.00	0.00	1.14	1.14	-6.81	-1.13
Night Wakings	-1.59	-3.92	0.00	-3.17	1.58	-3.17	1.58
Parasomnias	-2.94	-4.76	0.00	1.00	0.00	-1.96	0.98
Sleep-Disordered Breathing	0.00	-4.55	2.27	0.00	0.00	2.27	0.00
Daytime Sleepiness	0.00	-1.20	-1.80	-1.80	-2.40	-0.60	-0.60
Total Score	-2.92	-7.30	-0.73	-0.73	-2.19	-9.12	-1.46

Note. Negative values indicate a therapeutic reduction in the CSHQ scores, positive values indicate a countertherapeutic increase in CSHQ scores. There are no clinical cut-off scores for CSHQ subscales, therefore calculation of clinically substantive change was only possible for the Total Score.

Shading indicates the degree of reliable and clinical change in scores from pre- to post-treatment: = clinical deterioration; = reliable deterioration; = non-reliable change; = reliable improvement; = clinically substantive improvement.

SSR. Pre- and post-treatment scores for each SSR domain are presented in Table 7.3. The SSR does not possess a clinical cut-off score, and standardised data was not available so reliable and clinical change could not be assessed. Nevertheless, Table 7.3 shows all participants reported a large reduction in their SSR Total Score (mean reduction = -11), with Finn and Scott's post-treatment score close to the minimum score of 23.

Table 7.3. *Sleep Self Report (SSR) Pre- and Post-treatment Scores*

SSR Scales	Finn		Scott		Eve		Maximum
	Pre	Post	Pre	Post	Pre	Post	
Bedtime Resistance	4	2	2	2	4	4	6
Sleep Onset Delay	3	2	2	1	2	1	3
Sleep Duration	2	1	1	1	2	2	3
Sleep Anxiety	4	4	4	2	3	3	6
Night Wakings	3	2	4	2	4	3	6
Daytime Sleepiness	2	1	1	1	3	2	3
Total Score	43	28	38	25	41	36	69

CSC. As indicated by Table 7.4, all five children experienced a therapeutic reduction in their total Children's Sleep Comic (CSC) score. For Finn, Scott, and Eve this represented a reliable and clinically substantive improvement, with post-treatment scores falling below the severe sleep problems cut-off. The mean difference between participants pre- and post-treatment score on the CSC was -4.2.

Table 7.4. *Children's Sleep Comic (CSC) Standardised Change Scores with Reliable and Clinical Change Shaded*

	Seth	Finn	Ben	Scott	Eve
Total Score	-0.50	-2.48	-1.49	-4.46	-1.50

Note. Negative values indicate a therapeutic reduction in CSC scores.
Shading indicates the degree of reliable and clinical change in scores from pre- to post-treatment: ■ = clinical deterioration; ■ = reliable deterioration; □ = non-reliable change; ■ = reliable improvement; ■ = clinically substantive improvement.

ASWS-R and ASHS. John and Isaac's post-treatment ASWS-R (see Table 7.5) and ASHS scores were not reliably different from their pre-test scores. John and Isaac's standardised change scores for the ASHS were 0.85 and 0.70 respectively. Given their pre-treatment scores approached the maximum of 6 (4 - 5.5) on both scales, a ceiling effect may have prevented significant change. Additionally, as the ASHS and ASWS-R evaluate each sleep domain by calculating an average score across items, very large changes to raw scores would have been required to significantly alter the average score for each subscale and total score.

Table 7.5 *Adolescent Sleep Wake Scale Revised (ASWS-R) Standardised Change Scores with Reliable and Clinical Change Shaded*

ASWS-R Scales	John	Isaac
Falling Asleep and Reinitiating Sleep	-1.08	1.43
Returning to Wakefulness	0.26	0.00
Going to Bed	0.66	1.31
Total	-0.85	1.24

Note. Positive values indicate a therapeutic increase in ASWS-R scores. Negative values indicate a countertherapeutic reduction in ASWS-R scores.

Shading indicates the degree of reliable and clinical change in scores from pre- to post-treatment: ■ = clinical deterioration; ■ = reliable deterioration; □ = non-reliable change; ■ = reliable improvement; ■ = clinically substantive improvement.

Secondary Outcomes

Young persons' secondary outcomes.

GARS-3. As Figure 7.6 and Table 7.6 shows, Peter and John experienced reliable and clinically substantive reductions (from Requiring Very Substantial Support to Requiring Substantial Support) in ASD severity post-treatment, however, overall, the effect size for the GARS-3 ASD Index was small (d_{av} = -0.35; CLES = 67%) and d_{av} was not reliably different from 0 (95% CI [-0.88, 0.20]). No participant experienced a reliable deterioration in their ASD Index score.

Three participants experienced reliable reductions in GARS-3 subscale scores. For Peter this concerned the Restricted/Repetitive Behaviors and Emotional Responses subscales, for John the Social Interaction, Emotional Responses, and Cognitive Style subscales, and Eve the Maladaptive Speech subscale. Seth and Isaac experienced a reliable deterioration in Maladaptive Speech and Emotional Responses subscale scores respectively. The remaining four participants did not experience a reliable change in GARS-3 scores. Further, no participant experienced a reliable change on the Social Communication subscale score.

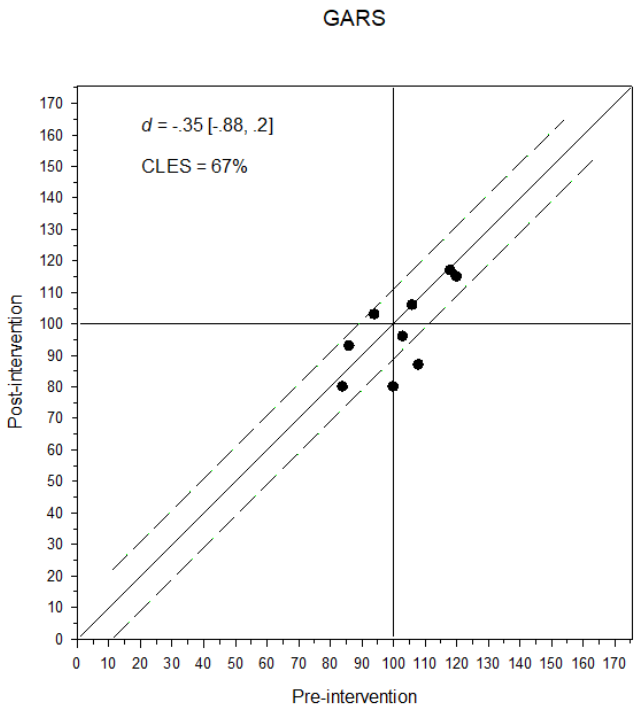

Figure 7.6. Modified Brinley plot showing change from pre- to post- intervention on the Gilliam Autism Rating Scale – Third Edition (GARS-3). *d* = Cohen's d_{av} effect size; CLES = common language effect size.

Table 7.6. *Gilliam Autism Rating Scale - Third Edition Standardised Change Scores with Reliable and Clinical Change Shaded*

	Niko	Eric	Peter	Seth	Finn	Scott	John	Isaac	Eve
Restricted/ Repetitive Behaviors	0.00	0.00	-3.54	-0.71	0.00	0.00	0.71	0.00	-1.45
Social Interaction	-1.42	0.00	-0.71	0.71	0.00	-0.71	-5.67	0.00	-0.71
Social Communication	0.71	-1.42	-0.71	1.42	-0.71	-0.71	-0.71	0.00	0.71
Emotional Responses	-1.42	-0.70	-2.13	0.00	-0.71	0.00	-2.13	2.12	-1.42
Cognitive Style	0.00	0.00	-1.42	0.00	-0.71	-0.71	-2.13	1.42	0.00
Maladaptive Speech	1.42	-0.70	-1.42	2.84	-1.42	0.00	0.00	0.00	-3.55
ASD Index	-0.18	0.00	-3.71	1.59	-0.71	-0.88	-3.53	1.23	-1.24

Note. Gilliam Autism Rating Scale - Third Edition = GARS-3; Negative values indicate a therapeutic reduction in GARS-3 scores. Positive values indicate a countertherapeutic increase in GARS-3 scores. There are no clinical cut-off values for the GARS-3 subscales, therefore degree of clinical change across the 6 subscales could not be assessed.

Shading indicates the degree of reliable and clinical change in scores from pre- to post-treatment: = clinical deterioration; = reliable deterioration; = non-reliable change; = reliable improvement; = clinically substantive improvement.

CBCL. As Table 7.7 indicates, seven out of ten parents reported a reliable improvement in CBCL problem behaviour scores from pre- to post-treatment, with all participants having reported clinically elevated levels of Total (behaviour) Problems pre-treatment. Two participants (Niko and Peter) had clinically substantive reductions in Total Problems, however this change was not reliable for Peter. All of the remaining eight participants stayed within the clinical range at post-treatment, but with three of these

participants having reliable reductions in total problem behaviours. Although Isaac did not have a reliable reduction in CBCL composite scale scores, parent responses yielded reliable reductions in three subscale scores, Attention Problems, Somatic Complaints, and Thought Problems, the latter of which were also clinically substantive improvements. Overall, the effect size was large for the CBCL Total scale (d_{av} = -0.84; CLES = 87%) and was reliably different from 0 (95% CI [-1.43, -0.22]).

Five participants were reported to experience a reliable improvement in internalising behaviour, which was clinically substantive for Finn. Four participants remained in the clinical range for internalising behaviour post-treatment and Peter remained in the non-clinical range. The effect size was medium for the CBCL Internalising scale (d_{av} = -0.57; CLES = 77%) and was reliably different from 0 (95% CI [-1.10, -0.02]).

Only Niko and Eve were reported to have a reliable improvement in externalising behaviour. For Eve, this change was clinically substantive. Peter and Eric remained in the non-clinical range for externalising behaviour and Seth and John remained in the clinical range post-treatment. Finn, Ben, and Scott's levels of externalising behaviour reduced from the Clinical to the Borderline range, however these changes were not within the bounds of reliable change. Overall, Cohen's *d* effect size was small for the CBCL Externalising scale (d_{av} = -0.36; CLES = 72%) and was not reliably different from 0 (95% CI [-0.76, 0.06]).

Table 7.7. *Child Behavior Checklist for Ages 6-18 Standardised Change Scores with Reliable and Clinical Change Shaded*

	Niko	Eric	Peter	Seth	Finn	Ben	Scott	John	Isaac	Eve
Internalising	0.00	-3.03	-0.88	-2.02	-5.05	-2.65	-0.44	0.88	-0.88	-10.38
Externalising	-2.08	0.42	-1.35	0.42	-1.67	-1.01	-1.68	0.34	1.35	-3.30
Total Problems	-3.10	-1.72	-1.85	-0.34	-1.21	-2.92	-2.92	0.31	-1.85	-8.00

Note. Negative values indicate a therapeutic reduction in internalising and externalising problem behaviours. Positive values indicate a countertherapeutic increase.

Shading indicates the degree of reliable and clinical change in scores from pre- to post-treatment: = clinical deterioration; = reliable deterioration; = non-reliable change; = reliable improvement; = clinically substantive improvement.

As Table 7.8 illustrates, no participants experienced reliable change in their overall CBCL competence score. However, parent responses yielded reliable and clinically substantive improvement in Niko and Eve's school competence specifically. The effect size was negligible for the CBCL Competence scale (d_{av} = -0.11, 95% CI [-0.31, 0.10]; CLES = 63%).

Table 7.8. *Child Behavior Checklist for Ages 6-18 Standardised Change Scores with Reliable and Clinical Change Shaded for the Competence subscale*

	Niko	Eric	Peter	Seth	Finn	Ben	Scott	John	Isaac	Eve
Competence	0.88	0.00	-1.09	0.29	-0.88	0.54	-0.54	-1.63	-1.36	0.82

Note. Positive values indicate a therapeutic increase in competence behaviours across social, activity, and school domains. Negative values indicate a countertherapeutic decrease in competence behaviours.

Shading indicates the degree of reliable and clinical change in scores from pre- to post-treatment: = clinical deterioration; = reliable deterioration; = non-reliable change; = reliable improvement; = clinically substantive improvement.

MASC-2 SR and PR. The effect sizes were large for the MASC-2 SR Total score (d_{av} = -0.89; CLES = 97%) and the mean difference between pre- and post-test scores was -31.2. The effect sizes were medium to large for the MASC-2 PR Total score (d_{av} = -0.70; CLES = 96%) and the mean difference between pre- and post-tests scores was -17.2.

Table 7.9 and 7.10 demonstrates that each participant and their parent reported a reduction in total anxiety scores on the MASC-2. Scott, Eve, John, and Isaac's self-reported Total Anxiety score was in the clinical range pre-treatment but at post-treatment Eve, John, and Isaac's self-reported anxiety had reduced to non-clinical levels. This change was only reliable for John and Isaac. Scott remained in the clinical range despite his total anxiety raw score reducing by 24, due to very high anxiety pre-treatment. Ben was the only participant in the non-clinical range for self-reported anxiety pre-treatment, and, despite a low baseline

level of anxiety, he too experienced a reliable reduction, with anxiety levels falling to the minimum possible level.

Based on parent report, four out of five participants (Finn, Scott, Eve, and John) experienced clinical levels of total anxiety pre-treatment. At post-treatment, three of these participants (Scott, John, and Eve) had experienced reliable improvements in total anxiety levels, although they remained in the clinical range. Despite Scott and Eve's Total Anxiety raw scores reducing by 23 and 30 points respectively, they remained within the clinical range due to very elevated levels of pre-treatment anxiety. Ben remained in the non-clinical range at post-treatment.

MASC-2 SR results revealed participants experienced reliable improvements across a range of anxiety dimensions, namely Separation Anxiety/ Phobias (one participant), General Anxiety Disorder (two participants), Social Anxiety (one participant), Obsessions and Compulsions (one participant), Physical Symptoms (four participants), and Harm Avoidance (one participant). Similarly, parent-report on the MASC yielded reliable reductions in Separation Anxiety/ Phobias for one participant, Social Anxiety (two participants), Obsessions and Compulsions (two participants), and Physical Symptoms (two participants).

Table 7.9. *Multidimensional Anxiety Scale for Children Self-report (MASC-2 SR) Standardised Change Scores with Reliable and Clinical Change Shaded*

MASC-2 SR Scales	Finn	Ben	Scott	Eve	John	Isaac
Separation Anxiety/ Phobias	-0.67	-2.03	-1.02	0.00	-0.67	0.00
General Anxiety Disorder	-3.26	-2.67	-0.30	-1.05	-0.74	-0.59
Social Anxiety Total	-2.19	-1.88	0.31	-0.57	-1.10	0.62
Humiliation/ Rejection	-2.63	-1.05	0.00	0.00	-1.05	0.52
Performance Fears	-0.87	-1.75	0.44	-0.80	0.00	0.43
Obsessions and Compulsions	1.67	-1.00	-1.00	-0.57	-3.01	-1.00
Physical Symptoms Total	-	-2.03	-3.39	-0.88	-3.39	-2.71
Panic	-	0.44	-3.54	-0.80	-3.39	-2.21
Tense/ Restless	0.00	-3.72	-1.06	-0.45	-1.60	-1.60
Harm Avoidance	0.00	-0.32	-2.25	-0.64	-0.96	-0.64
Total Score	-	-3.33	-3.08	-1.27	-6.67	-5.51

Note. Negative values indicate a therapeutic reduction in anxiety symptoms. Positive values indicate a countertherapeutic increase.

Shading indicates the degree of reliable and clinical change in scores from pre- to post-treatment: = clinical deterioration; = reliable deterioration; = non-reliable change; = reliable improvement; = clinically substantive improvement.

Table 7.10. *Multidimensional Anxiety Scale for Children Parent-report (MASC-2 PR) Standardised Change Scores with Reliable and Clinical Change Shaded*

MASC-2 PR Scales	Finn	Ben	Scott	Eve	John
Separation Anxiety/ Phobias	-0.36	-0.59	-3.04	-1.35	-1.44
General Anxiety Disorder	-0.93	-0.72	-0.62	-1.52	0.00
Social Anxiety Total	-1.05	-2.81	0.00	-2.34	-1.40
Humiliation/ Rejection	-0.59	-3.55	-0.59	-3.23	0.59
Performance Fears	-0.94	-0.94	0.47	-0.51	-2.36
Obsessions and Compulsions	0.00	0.48	-1.46	-2.17	-2.91
Physical Symptoms Total	0.40	-1.21	-4.45	-2.17	0.00
Panic	0.00	-0.57	-4.04	-1.26	0.00
Tense/ Restless	0.56	-1.12	-2.25	-1.92	0.00
Harm Avoidance	-0.69	0.69	-1.04	-1.39	-0.69
Total Score	-0.89	-1.64	-3.42	-4.35	-2.38

Note. Negative values indicate a therapeutic reduction in anxiety symptoms. Positive values indicate a countertherapeutic increase.

Shading indicates the degree of reliable and clinical change in scores from pre- to post-treatment: ■ = clinical deterioration; ■ = reliable deterioration; □ = non-reliable change; ■ = reliable improvement; ■ = clinically substantive improvement.

Parent secondary outcomes.

PSQI. As shown in Figure 7.7 and Table 7.11, seven of the fifteen parents included in this analysis were in the clinical range for sleep problems pre-treatment and two (Niko's father and John's mother) experienced a clinically substantive improvement in their own sleep, without this being reliable change for Niko's father. The majority (8/15) of parents were in the non-clinical range for sleep problems pre-treatment. Ben's father experienced a reliable and clinically substantive deterioration in his sleep and the remaining parents remained within the non-clinical range. The effect sizes were negligible for the PSQI (d_{av} = -0.19, 95% CI [-0.59, 0.22]; CLES = 60%).

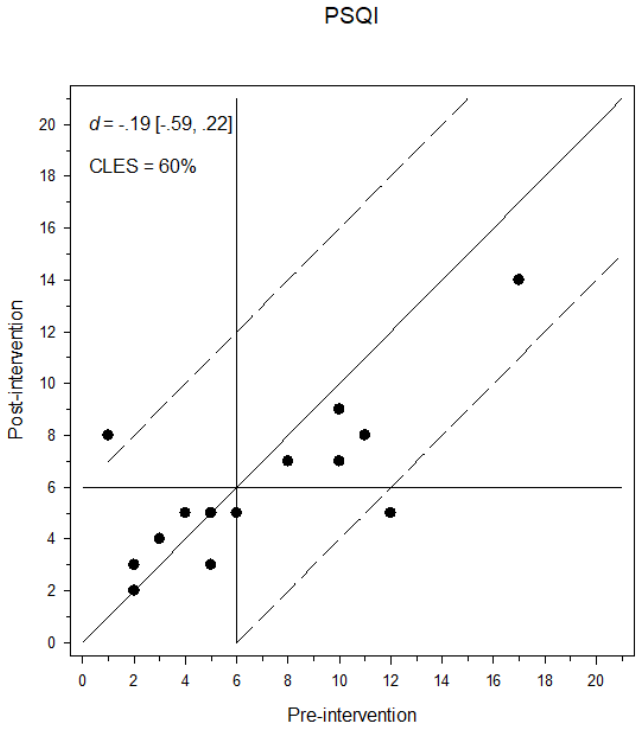

Figure 7.7. Modified Brinley plot showing change from pre- to post- intervention on the Pittsburgh Sleep Quality Index (PSQI). *d* = Cohen's d_{av} effect size; CLES = common language effect size.

Table 7.11. *Parent Pittsburgh Sleep Quality Index (PSQI) Standardised Change Scores with Reliable and Clinical Change Shaded*

	PSQI Global Score
Niko	
Father	-0.34
Eric	
Mother	-1.01
Peter	
Mother	0.34
Father	0.00
Seth	
Mother	-0.35
Father	-
Finn	
Mother	-0.67
Father	0.00
Ben	
Mother	0.34
Father	2.36
Scott	
Mother	-1.01
Father	-
Eve	
Mother	-0.34
Father	0.00
John	
Mother	-2.36
Isaac	
Mother	-1.01
Father	0.34

Note. Negative values indicate a therapeutic decrease in PSQI scores. Positive values indicate a countertherapeutic increase in PSQI scores.

Shading indicates the degree of reliable and clinical change in scores from pre- to post-treatment: = clinical deterioration; = reliable deterioration; = non-reliable change; = reliable improvement; = clinically substantive improvement.

DASS-21: Depression. Figure 7.8 and Table 7.12 show there was a reliable improvement in Depression subscale scores for four out of nineteen parents, and for three of these parents this was clinically substantive. Two mothers also experienced a reliable deterioration in their Depression subscale score. This was clinically substantive for one parent, however the other parent remained within the non-clinical range. Effect sizes were small for the DASS-21 Depression scale (d_{av} = -0.34 95% CI [-0.90, 0.22]; CLES = 61%) and not reliably different from 0.

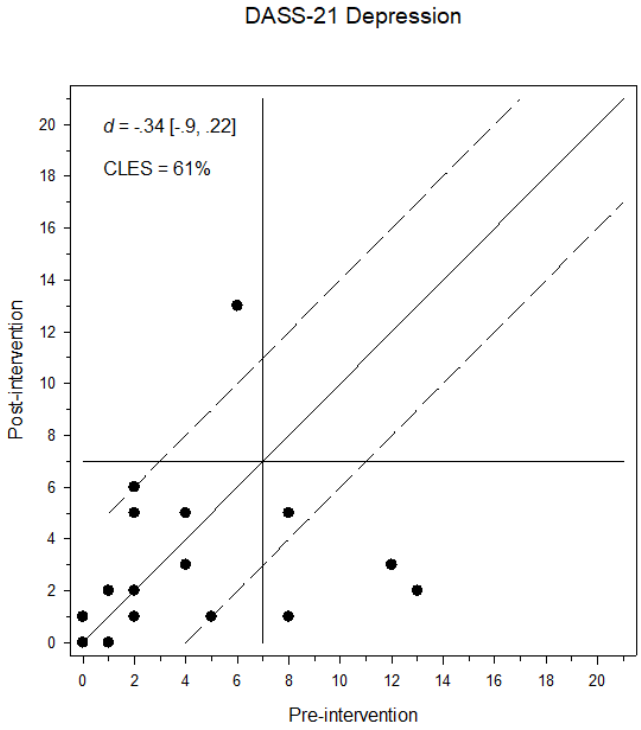

Figure 7.8. Modified Brinley plot showing change from pre- to post- intervention on the Depression Anxiety Stress Scales 21 (DASS-21) Depression subscale. *d* = Cohen's d_{av} effect size; CLES = common language effect size.

DASS-21: Anxiety. Four parents experienced a clinically substantive improvement in anxiety levels, but one did not reach the threshold of reliable change (see Figure 7.9; see Table 7.12). Fourteen out of the remaining fifteen parents remained within the non-clinical range and one (Eve's father) experienced a reliable and clinically substantive increase in anxiety. He was the only parent whose responses yielded an Anxiety scale score in the clinical range. The effect size was medium for the DASS-21 Anxiety scale (d_{av} = -0.55; CLES = 68%) and not reliably different from 0 (95% CI [-1.10, 0.01]).

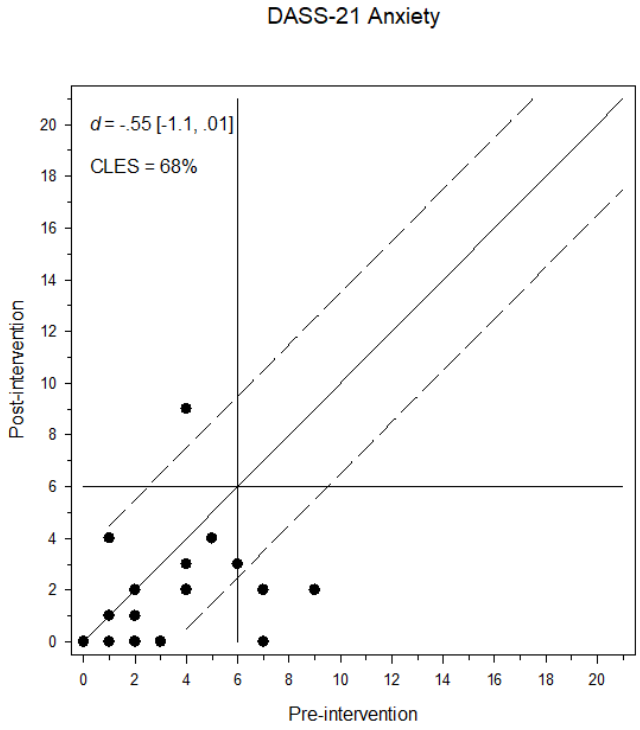

Figure 7.9. Modified Brinley plot showing change from pre- to post- intervention on the Depression Anxiety Stress Scales 21 (DASS-21) Anxiety subscale. *d* = Cohen's d_{av} effect size; CLES = common language effect size.

DASS-21: Stress. Four parents experienced a clinically substantive improvement in stress, two of which exceeded the RCI (see Figure 7.10; see Table 7.12). Three parents remained within the clinical range, although one experienced a reliable improvement (Blair's mother). Eleven parents remained within the non-clinical range and one parent (Ben's mother) experienced a reliable and clinically substantive deterioration in stress. The effect size was small for the DASS-21 Stress scale (d_{av} = -0.24; CLES = 62%) and not reliably different from 0 (95% CI [-0.58, 0.11]).

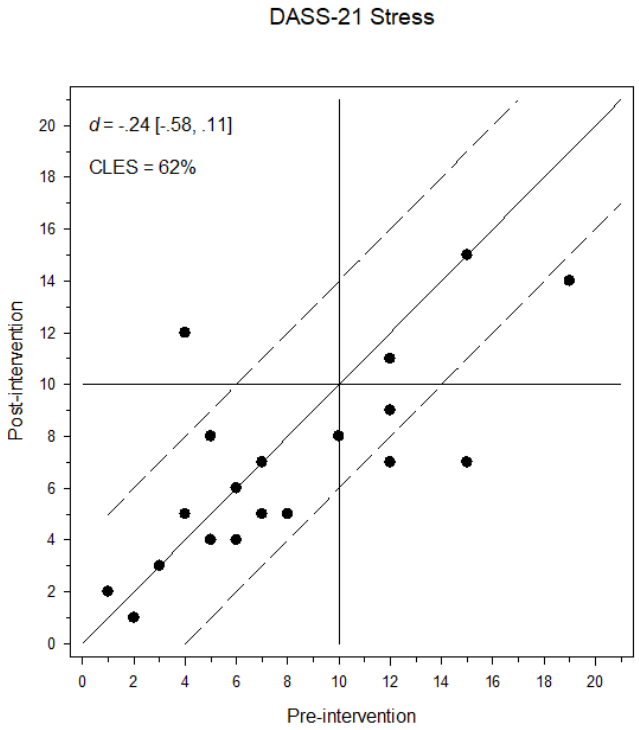

Figure 7.10. Modified Brinley plot showing change from pre- to post- intervention on the Depression Anxiety Stress Scales 21 (DASS-21) Stress subscale. *d* = Cohen's d_{av} effect size; CLES = common language effect size.

DASS-21: Total. The effect size was small for the DASS-21 Total scale (d_{av} = -0.38; CLES = 66%) and not reliably different from 0 (95% CI [-0.80, 0.06]; see Figure 7.11; see Table 7.12). Five parents experienced reliable improvement in DASS-21 Total scores and two experienced reliable deterioration. Summarising DASS-21 scores overall, they fell within the Normal range pre- and post-treatment for Peter's parents, Finn's parents, Niko's father, Ben's father and Isaac's father. Blair's father's Depression and Anxiety scores were in the Normal to Mild range both pre and post-treatment. Consequently, floor effects limited capacity for reliable change in DASS-21 scores for these eight parents. Conversely, more mothers experienced high levels of depression, anxiety, and stress pre-treatment, which were reliably improved at post-treatment. Interestingly, maternal wellbeing was also more likely to deteriorate post-treatment than paternal wellbeing.

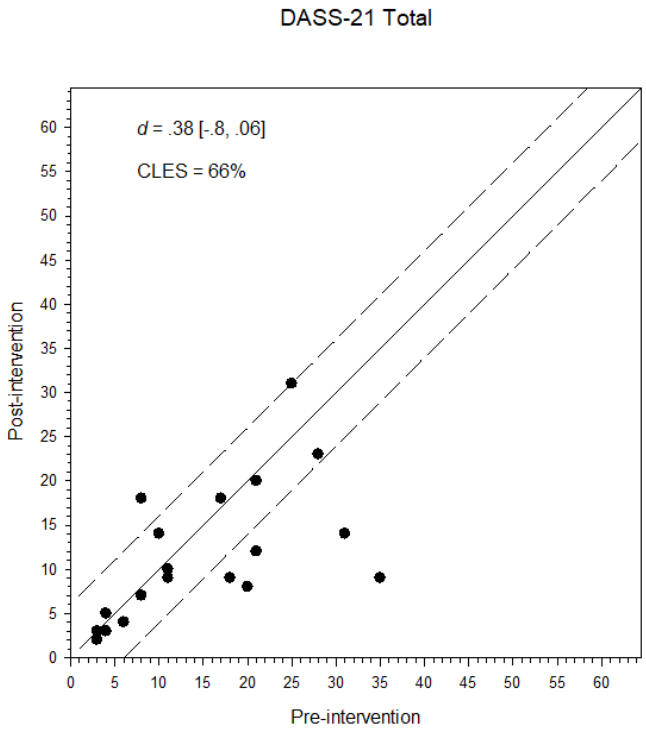

Figure 7.11. Modified Brinley plot showing change from pre- to post- intervention on the Depression Anxiety Stress Scales 21 (DASS-21) Total scale score. *d* = Cohen's d_{av} effect size; CLES = common language effect size.

Table 7.12. *Parent Depression Anxiety Stress Scales 21 Standardised Change Scores with Reliable and Clinical Change Shaded*

	Depression	Anxiety	Stress	Total
Blair				
Mother	-1.58	1.70	-2.66	-1.39
Father	0.53	-0.56	-0.53	-0.28
Niko				
Father	-0.53	-0.56	0.53	-0.28
Eric				
Mother	-2.11	-1.69	-1.06	-2.49
Peter				
Mother	-0.53	0.00	0.00	-0.28
Father	-0.53	0.00	-0.53	-0.55
Seth				
Mother	2.11	-1.69	1.60	1.11
Father	-4.75	-2.82	-1.60	-4.70
Finn				
Mother	-0.53	0.00	0.00	-0.28
Father	-0.53	1.13	-1.06	-0.28
Ben				
Mother	1.58	-0.56	4.26	2.76
Father	0.00	0.00	0.53	0.28
Scott				
Mother	0.00	-3.95	-1.06	-2.49
Father	-0.53	-0.56	0.00	-0.55
Eve				
Mother	-3.69	-1.13	-1.60	-3.32
Father	0.53	3.95	-2.65	0.27
John				
Mother	-5.80	-3.95	-4.26	-7.19
Isaac				
Mother	3.69	-0.56	0.00	1.66
Father	0.53	0.00	-0.53	0.00

Note. Depression Anxiety Stress Scales-21 = DASS-21; Negative values indicate a therapeutic decrease in DASS-21 scores. Positive values indicate a countertherapeutic increase in DASS-21 scores.

Shading indicates the degree of reliable and clinical change in scores from pre- to post-treatment: = clinical deterioration; = reliable deterioration; = non-reliable change; = reliable improvement; = clinically substantive improvement.

RQI. Figure 7.12 and Table 7.13 show Seth's father experienced a clinically substantive improvement in parental relationship quality, increasing from a score indicative of relationship distress, to relationship satisfaction. All other parents remained within the non-clinical range for relationship quality, indicating ongoing relationship satisfaction. The effect size was medium for the RQI (d_{av} = 0.50) but not reliably different from 0 (95% CI [-0.04, 1.03]; CLES = 74%).

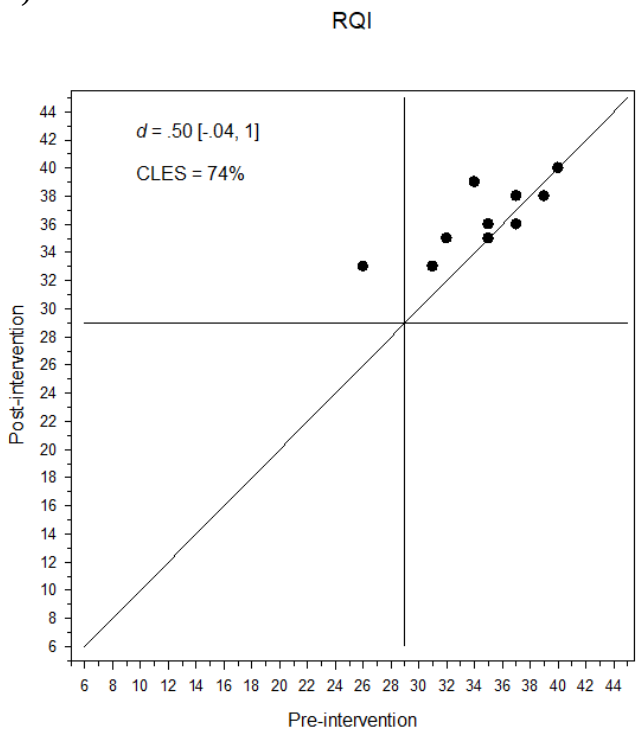

Figure 7.12. Modified Brinley plot showing change from pre- to post- intervention on the Relationship Quality Index (RQI). *d* = Cohen's d_{av} effect size; CLES = common language effect size.

Table 7.13. *Parent Relationship Quality Index Scores Pre- and Post-Treatment*

	Seth		Finn		Ben		Scott		Eve	
	Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post
Mother	37	36	40	40	39	38	31	33	35	35
Father	26	33	34	39	37	38	35	36	32	35

Note. Pre = pre-treatment. Post = post-treatment. Higher scores are indicative of higher relationship quality. Scores below 27 are indicative of relationship distress.

Shading indicates the degree of reliable and clinical change in scores from pre- to post-treatment: = clinical deterioration; = reliable deterioration; = non-reliable change; = reliable improvement; = clinically substantive improvement.

Discussion

It is vital to a comprehensive understanding of the nature of ASD and its treatment on diagnosed children, siblings, parents and other family members, to understand the association between sleep problems and daytime functioning and wellbeing, particularly given individuals with ASD may already experience significant difficulties in these areas. Given the strong association between paediatric sleep quality and the overall wellbeing of young people and their whānau/family, it was hypothesised that resolving participant's sleep would also improve the wellbeing of them and their parents. Overall, diary-based and psychometric measures of sleep indicated that participant sleep disturbance improved to varying degrees from pre- to post-treatment. Secondary benefits included improvements in total problem behaviour and internalising behaviour, as measured by the CBCL, as well as improvements in anxiety as measured by the MASC-2 SR and PR. However, there was minimal change in GARS-3 ASD symptom severity or in externalising and competent behaviour assessed by the CBCL. Further, there was little change in parent wellbeing as measured by the DASS-21, PSQI, and RQI, although most parents were already in the non-clinical range for these variables pre-treatment.

Sleep Outcomes

Overall, questionnaire results indicated participant sleep disturbance reduced from pre- to post-treatment and parent- and self-reported results tended to correspond with one another. Despite there being a reliable effect for the CSHQ, five out of seven participants remained in the clinical range for sleep disturbance at post-treatment. This is in contrast to parent-reported sleep diaries and VSG which indicated most participants experienced significant reductions or elimination of sleep disturbances. Sensitivity and specificity ratings for the CSHQ clinical cut-off demonstrate this value will likely capture true clinical cases 80% of the time and capture true non-clinical cases only 72% of the time (Owens, Spirito, & McGuinn, 2000). Consequently, 28% of participants who did not have clinical levels of sleep disturbance may have been falsely identified as being in the clinical range. Research shows objective sleep measures correlate more strongly with sleep diaries than they do with questionnaires (Acebo et al., 2005). As sleep diaries gather specific numerical data continuously, they are likely to be more accurate than questionnaires which are global estimations of sleep disturbance and do not necessarily capture intraindividual variability (Acebo et al., 2005).

Secondary Outcomes - Young Person

GARS-3: Autism symptoms. The GARS- 3 confirmed that participants had severe levels of ASD pre-treatment. In general, findings suggest that participants did not experience reliable reductions in ASD symptom severity following a sleep intervention. Few studies have examined ASD symptoms pre- and post-treatment for sleep disturbance. In contrast to the current study, Reed et al. (2009), Malow et al. (2014), and McCrae et al. (2019) found significant improvements in repetitive behaviours and stereotypy in children aged 2 to 12 years following parent sleep education programmes or CBT-I for insomnia. However, similar to the current study, Loring et al. (2018) found that an adolescent- and parent-implemented behavioural sleep intervention did not result in reduced repetitive motor behaviour, or restricted/ritualistic behaviour, and insistence on sameness. A possible reason for this is that most correlational studies examining the relationship between ASD symptoms and sleep disturbance have included young, non-verbal children (Loring et al., 2018). Such children may be more likely to engage in sleep-interfering motor behaviour than verbal adolescents with ASD (Loring et al., 2018). Consequently, such behaviour is likely to be addressed and reduced during the course of a targeted sleep intervention. Within the current study, FBA revealed ASD symptoms measured by the GARS-3 (e.g., Maladaptive Speech, "Speech is abnormal in tone, volume, or rate"; Cognitive Style, "Displays superior knowledge or skill in specific subjects") were rarely directly implicated within participant sleep disturbance and not specifically targeted, which may aid in explaining the lack of association.

CBCL: Challenging Behaviour. Given the relationship between sleep and executive functions, such as impulse control, it was hypothesised that improved sleep may directly contribute toward better emotional and behavioural regulation, resulting in less challenging behaviour. However, overall, there was no reliable difference in externalising behaviour pre- and post-treatment. Externalising behaviour that occurs in the context of bedtime resistance (e.g., tantrums) is less common among older children and adolescents than toddlers and young children with ASD (Goldman et al., 2012). Consequently, such behaviour was rarely directly addressed during intervention. Further, as interventions primarily consisted of less restrictive and/or young person-implemented components (e.g., psychoeducation versus extinction), parents did not necessarily acquire general behaviour management skills that could be applied to challenging behaviour during the day. These factors may have limited the extent of likely change in externalising behaviour following intervention.

Overall, there was a reliable improvement in the CBCL Total Problem scale from pre- to post-treatment. This is consistent with other studies which have found improvements in internalising behaviour, externalising behaviour, irritability, lethargy, hyperactivity, and attention problems following behavioural treatment of sleep problems (Malow et al.,2014; McCrae et al., 2019; Moon et al., 2011). In contrast, Moss et al. (2014) did not find any reliable change in participants' Developmental Behaviour Checklist (e.g., emotional and behavioural disturbance, Einfeld and Tonge, 2002) scores following a parent education programme. The authors suggest problem behaviours measured by the checklist were likely reflective of the children's disability and therefore would be less amenable to change in response to a brief intervention (Moss et al., 2014).

The CBCL Total Problems scale consists of internalising and externalising behaviour scales as well as Social Problems, Thought Problems, Attention Problems and Other Problems scales. Most participants experienced reliable reductions in Social, Thought, and Attention Problems post-treatment. Such results are in line with Phung and Goldberg's (2017) study which showed sleep disturbance can lead to further impairment in the social communication skills of young people with ASD. Further, as lack of sleep contributes to daytime sleepiness and reduced alertness, it is perhaps not surprising that better sleep would improve neurological functions, such as attentional capacities (Bernier, Carlson, Bordeleau, & Carrier, 2010; Dahl, 1996; Sadeh, Gruber, & Radiv, 2002; Malow et al., 2014; Turner & Johnson, 2013). Of note, the CBCL Thought Problems subscale contains three items relating to sleep. Therefore, improvements in this domain may reflect improved sleep as opposed to a reduction in other thought problems (e.g., "can't get mind off thoughts").

CBCL: Competence (activities, social, academic). Despite participants experiencing significant improvements in overall problem behaviour, this did not lead to a reliable increase in Competence scale scores. In fact, no participant experienced a reliable change on this scale. Only Niko's father's responses yielded a reliable and clinically substantive report of improvement in his child's School Competence post-treatment. Prior to intervention, Niko sometimes stayed home from school following a poor night's sleep but in the course of intervention Niko's school attendance improved. In addition, his alertness, attentiveness, and engagement in class activities likely increased, potentially contributing to an improvement in academic abilities. Interestingly, anecdotal parent report and responses on the CBCL Social Problem scale indicated participant's social communication had improved, however, this was

not reflected by the CBCL Social Competence scale or the GARS-3 Social Interactions and Social Communication scales. Few studies have examined improvement in children's strengths and desired behaviour post-treatment and instead have focused on reductions in behavioural problems. However, preliminary research suggests children with ASD can experience a significant improvement in quality of life post-treatment (Malow et al., 2014), as measured by The Parent Proxy-Report of the Pediatric Quality of Life Inventory (Varni, Limbers, & Burwinkle, 2007).

Internalising behaviour (CBCL) and anxiety (MASC-2). Parent responses on the CBCL post-treatment yielded significant improvements in participants internalising behaviour, including Anxious/Depressed behaviour (e.g., "nervous, high-strung, or tense", "feels worthless or inferior"), Withdrawn/Depressed behaviour (e.g., "refuses to talk", "unhappy, sad, or depressed"), and Somatic Complaints (e.g., "headaches", "stomach aches"). These findings were corroborated by MASC-2 results which revealed there were significant reductions in both parent-reported and self-report anxiety. Of note, the CBCL internalising behaviour scale includes items which may be a direct consequence of improved sleep (e.g., "underactive, slow moving, or lacks energy", "nightmares"), as opposed to mood problems, thus contributing to the significance of the relationship. However, recent research by Loring et al. (2018) and Malow et al. (2014) obtained similar improvements in internalising behaviour using additional measures.

In the current study, sleep interventions directly countered hyperarousal (e.g., faded bedtime) as well as taught skills (e.g., relaxation techniques), applicable throughout the day and night, which may have facilitated participant's regulatory capabilities. Individual results highlight the impact of intervention on participant anxiety. According to the MASC-2 SR and/or PR there was a reliable reduction in Separation Anxiety/ Phobias for Ben and Scott both of whom learnt to initiate sleep without seeking parent comfort or presence respectively. Ben, Scott, Eve, John, and Isaac all experienced a reliable reduction in Physical Symptoms of anxiety. Eve and John experienced a reliable reduction on the Obsessions and Compulsions scale, indicative of a reduction in intrusive thoughts. Scott experienced a reliable and clinically substantive reduction in Harm Avoidance, suggesting increased confidence to tolerate anxiety provoking situations (e.g., falling asleep independently) without engaging in safety behaviours. Finally, Finn, Ben, and Eve all experienced a reliable reduction in their Social Anxiety scale score. Such findings indicate sleep interventions can have a significant

impact on the internalising behaviour of young people with ASD. The bidirectional relationship between sleep and mood problems in young people with ASD, means sleep interventions could be a suitable first step in addressing depression and anxiety in this population (White et al., 2018).

Secondary Outcomes- Parent

PSQI: Sleep. In general, parents did not experience a reliable improvement in their own sleep from pre-treatment to post-treatment according to the PSQI. Existing literature suggests parents of children with sleep disturbance tend to experience sleep problems also (Lopez-Wagner et al., 2008). One purported reason for this relationship is that the child's sleep-interfering behaviour may interrupt or prevent parent sleep (Lopez-Wagner et al., 2008), however, this may not hold true for older children and adolescents who, as in this study, seem to engage more in covert sleep-interfering behaviour which is not disruptive to the sleep of other household members. It is unclear why Ben's father experienced a clinically substantive deterioration in his PSQI sleep score, given Ben learnt to independently reinitiate sleep upon waking during the night.

McCrae et al. (2019) found parents in their study experienced improvements in both actigraphic and diary measured sleep following eight CBT-I sessions delivered to them and their child (aged 6 to 12 years). Actigraphic data showed that, on average, parents fell asleep 12 minutes faster, woke for 21 minutes less during the night, and experienced a 4% increase in SE post treatment. As parents in the current study had a less pronounced role in intervention, they may not have benefited from psychoeducation and sleep-conducive strategies (e.g., relaxation strategies) taught to young people. This is in contrast to McCrae et al's (2019) study where parents played a more active role. Additionally, as the PSQI is a global measure of parent sleep it may have been less sensitive to differences in individual sleep variables (e.g., SOL, duration of WASO), as supported by its low convergent validity with PSG data.

DASS-21: Depression, anxiety, and stress symptoms. Existing research into associations between sleep disturbance in children with ASD and parental stress and poor mental health (Duarte, Bordin, Yazigi, & Mooney, 2005; Hodge et al., 2013; Hoffman et al., 2008; Johnson et al., 2018; Tilford et al., 2015) suggests that sleep disturbance is correlated with impaired parental wellbeing. However, most of this research is cross-sectional and does

not illustrate whether changes to child sleep affect parental mental health (Martin et al., 2019). Preliminary research by Moss et al. (2014) indicated parents experienced a clinically substantive reduction in their stress (according to the Parenting Stress Index, Abidin 1995) following a behavioural sleep intervention.

In the current study, there was no reliable improvement in overall parent mental health, as indicated by the DASS-21 Total score as well as the individual subscales. This is probably because most parents were in the non-clinical range for Depression, Anxiety, and Stress already, therefore limiting the extent of reliable improvement possible. There tended to be more clinical and/or reliable change in scores for parents who indicated severe levels of Depression, Anxiety, and Stress pre-treatment. For example, four out of nineteen parents were in the clinical range for Depression and/or Anxiety pre-treatment and at post-treatment this had reduced to one out of sixteen. Further, seven out of nineteen parents were in the clinical range for Stress pre-treatment, which reduced to four at post-treatment.

There was a reliable deterioration in scale scores for three mothers and one father at post-treatment. Although there is a strong relationship between child sleep and parent mental health, other factors may have also contributed to mental health outcomes. Parents of children with neurodevelopmental disorders often face chronic additional challenges, such as managing a range of complex behavioural difficulties (e.g., self-injurious behaviour) and supporting independent functioning (e.g., assist with hygiene tasks, Lee, 2009; Schnabel et al., 2020). Parents whose mental health deteriorated were also experiencing additional stressors, such as relationship breakdowns, moving to a new house, and undergoing assessment for other child psychological issues. Further, it is possible that stress associated with the intervention may have contributed to worse mental health outcomes. Seth and Ben's mothers, who experienced a reliable deterioration in Depression and a reliable deterioration in Stress and Total score respectively, indicated the least satisfaction with intervention procedures according to the TARF-R.

Most of the existing research in this area has focussed on maternal wellbeing (Martin et al., 2019), however, the current study investigated the impact of a behavioural sleep intervention on both parents. Overall, fathers tended to report lower levels of depression, anxiety, and stress symptoms pre-treatment and subsequently experienced less change post-treatment. By contrast, mothers were more likely to experience mental health difficulties pre-treatment and thus experienced greater change. Within the current study, mothers tended to

take primary responsibility for their child's sleep management and consequently may have been more likely to be affected by the intervention. Kirkpatrick et al. (2019) found inequity regarding child sleep management contributed to maternal anxiety and stress, as well as marital discord.

RQI: Parent marital relationship. In general, research suggests parents of children with ASD have lower marital satisfaction than parents of typically developing children (Benson & Kersh, 20011; Lee, 2009; Rodrique, Morgan, & Geffken, 1990). This is thought to be related to additional stress and childcare demands, as well as conflict regarding management of complex behavioural difficulties, such as sleep disturbance (Benson & Kersh, 2011). Consequently, it was hypothesised that intervention would likely improve parent relationship quality. Although most parents experienced an improvement in overall relationship quality, this change was not reliably greater than measurement error. Unexpectedly, a ceiling effect may have limited clinically significant improvement. All parents were in the non-clinical range for relationship satisfaction pre-treatment, apart from one parent who experienced a clinically substantive improvement post-treatment. Interestingly, in Sanberg et al's (2018) study, a ceiling effect also limited improvement in marital relationship quality following a behavioural sleep intervention for three 4- to 8-year olds with ASD. In Risdal and Singer's (2004) meta-analysis of marital relationship quality among parents of children with disabilities, they found low rates of marital discord when compared to parents of typically developing children. They suggest the presence of a family member with a disability does not inevitably contribute to negative outcomes but that in some cases it may in fact strengthen families and build resilience (Risdal & Singer, 2004).

Limitations

There are a number of limitations which need to be considered when interpreting the findings in this chapter. Firstly, the small sample size limits the generality of the conclusions drawn. Secondly, psychometric data was obtained only once soon after treatment had concluded and so the durability of secondary outcomes, whether by way of improvement or worsening, is unknown. Further, there was substantial variation in the length of time post-treatment that questionnaires were completed. Results obtained up to 6 weeks post-treatment may not accurately reflect participant and/or parent wellbeing immediately following intervention, particularly if effective treatment was not being adhered to at the later time. Alternatively, long-term data collection would enable investigation of changes that may only

occur following a sustained period of improved sleep, such as increased parental capacity to address other challenging behaviours. Future research could assess wellbeing variables immediately post-treatment as well as at short-term and long-term follow-up to establish the maintenance of effects.

Another limitation of these findings, given the unique nature of each participant's sleep disturbance, is that it is unknown which target behaviour improvements relate most strongly to secondary outcomes for parents and young people. In particular, some participants experienced improvements, such as learning to initiate sleep independently, that may not necessarily have contributed to better sleep quality or longer duration. Therefore, it cannot be specifically asserted that longer sleep duration relates to improvements reported in the current study (e.g., reduced anxiety).

A number of limitations relate specifically to the use of psychometric measures as a means of capturing behaviour change. Although sleep outcomes were assessed via both diaries and questionnaires, secondary outcomes were only assessed via questionnaires which are at greater risk of bias. The current study was particularly vulnerable to response bias given participants were not blind to the intention of the intervention (e.g., to improve sleep) or the rationale for completing questionnaires pre- and post-treatment (e.g., to evaluate change over time). Such knowledge may have inadvertently affected their responses. Further, the retrospective nature of questionnaires may have contributed to recall bias, such as differential remembering of more salient events, potentially masking small but real change. Future studies could consider triangulating information regarding secondary outcomes through observation, interviews, and/or formal testing (e.g., neurocognitive assessments).

Importantly, a number of the questionnaires had not been validated for use with young people with ASD, compromising their validity and reliability. Concordantly, normed information was not available from populations more representative of the current study. Cognitive abilities may also have inhibited participants' ability to self-monitor and accurately assess their own thoughts, feelings, and behaviour (Kreslins et al., 2015), this being particularly true for those with an ID. Despite altering items to facilitate comprehension, literal interpretation of questionnaire items was still problematic at times. For example, one participant responded negatively to the SSR item "Do you fall asleep in about 20 minutes?", as his SOL was not exactly 20 minutes each night. In particular, research has found small correlations between scores on the CSHQ versus SSR, potentially indicating inaccurate

responses from either party (e.g., the child is unable to evaluate SOL length, or parents are unaware of the extent of the sleep problem, Owens, Spirito, McGuinn & Nobile, 2000). Therefore, the measured data should be interpreted with caution.

Construct validity scores suggested that the chosen questionnaires could accurately measure the relevant psychological phenomena (assuming the constructs themselves had been accurately defined). Indeed, it is not surprising that there were reliable changes in both the CBCL Internalising behaviour and MASC-2 scores. However, differences between video, parent-report, and self-report sleep diary data compared with data obtained from the CSHQ, ASHS, and ASWS-R suggest such measures may evaluate different aspects of the same construct, or perhaps are not valid for use with the current sample.

Finally, it is important to note that reliable and/or clinically substantive change in some psychological variables is unlikely for individuals with lifelong disorders, as true of participants in the current study. Resolving sleep disturbance cannot eliminate clinical problems related to the presence of a neurodevelopmental disorder. Consequently, it is notable that participants experienced significant improvements in internalising and other challenging behaviours associated with ASD post-treatment. Further, secondary outcomes such as improved cognitive functioning, may take a long time to come into effect and therefore would not have been detected within this relatively short-term study. These factors may explain the reason for the lack of change in some of the secondary measures.

Conclusion

There are known to be strong associations between sleep and wellbeing for children, adolescents, and adults. This study illustrates that young people with ASD can experience significant improvements in internalising behaviour, such as anxiety and depression symptoms, as well as attention, thought, and social problems, following an FBA-informed sleep intervention. However, minimal change in ASD symptoms, externalising behaviour, or competence in social, school, and extra-curricular activities occurred in the short-term. In general, most parents within the current study experienced non-clinical levels of depression, anxiety, and stress symptoms, as well as marital discord, and sleep problems pre-treatment, with limited change post-treatment. These findings demonstrate improvement in sleep is not necessarily consistent with improved wellbeing, highlighting the complex and bidirectional nature of these interactions. The relationship between sleep problems in people with ASD and

child and parent wellbeing has yet to be determined (Malow et al., 2014, Martin et al., 2019). Future research is necessary to investigate the mechanisms underlying these relationships. This should involve consideration of the interactions within and between the young person's bioecological systems, such as the impact on the wider family (e.g., siblings). This is critical to optimising treatments for parent and child sleep, daytime functioning, and mental health (Martin et al., 2019).

Chapter 8

General Discussion

Since the 1960's, numerous studies have demonstrated the efficacy of behaviourally-based approaches to the treatment of paediatric sleep disturbance. However, a paucity of literature has examined the application of FBA-informed sleep interventions with verbal, older children and adolescents with ASD, or actively included the young person in the assessment and treatment process. Thus the primary aims of the current research were threefold: 1) to investigate the effectiveness of FBA-informed young person- and parent-implemented sleep treatment for verbal young people with ASD; 2) to explore whether FBA-informed assessment and treatment approaches are acceptable to young people and their parents; and 3) to assess change in participant and parent wellbeing post-treatment.

The three empirical studies reported in this thesis included 12 young people (11 males and 1 female aged 9 to 15 years) with ASD. The outcomes of this research highlight the feasibility, social validity, and effectiveness of FBA-informed sleep interventions involving input from the young people themselves. Study 1 (pilot study), demonstrated the feasibility and potential benefit of including young people with ASD as active intervention agents (alongside their parents), within comprehensive, individualised sleep treatments. Study 2 extended these findings and illustrated sleep disturbance could be treated via young person-implemented intervention alone in two cases, while some form of parent-implemented treatment (e.g., reinforcement) was necessary in the remaining 6/8 cases. Study 3 showed ASD-related sleep disturbance can be addressed via minimally sufficient, antecedent-based interventions only. Finally, data obtained across the three studies revealed while there was some improvement in participant anxiety and behavioural difficulties (e.g., inattentiveness, social problems) post-treatment, there was no reliable change in ASD severity and externalising behaviour, or to parent sleep, wellbeing, and relationship quality. This chapter presents the overall findings relating to each research question, the implications and limitations of these findings, and recommendations for future research.

The Effectiveness of FBA-informed Young Person- and Parent- implemented Sleep Interventions

All 12 participants demonstrated an improvement in target sleep variables from baseline to treatment. Moreover, sleep problems were no longer of concern for 10 participants at short- and long-term follow-up, as indicated by parent-report, self-report, and/or VSG. For the remaining two participants, sleep disturbance returned to baseline levels or deteriorated at follow-up. Interestingly, for each of these participants, this deterioration corresponded with a reduction in treatment fidelity. In three cases, where a secondary collection of long-term follow-up data was able to be undertaken, sleep disturbances remained resolved 18 to 24 months post-treatment. Collectively, the present research suggests FBA-informed sleep interventions, implemented by the young person in part, can effectively treat a range of dyssomnias (e.g., CCs, SOL, frequent and/or extended NWs, EWs, unwanted co-sleeping, and excessive daytime sleepiness) in older children and adolescents with ASD. Importantly, intervention was effective for all participants although they were diverse in communicative abilities, comorbid physical and mental health issues, nationality, socioeconomic status, and family composition.

FBA revealed a range of case-specific antecedent and consequence variables appeared to contribute to each participant's sleep disturbance, including, poor sleep hygiene, poor stimulus control, lack of physiological sleep pressure, sleep-competing dependencies, cognitive and physiological hyperarousal, sleep environment discomfort, exposure to light, and external noise. Participant behaviour that interfered with sleep was often multi-functional, relating to a combination of the following functions: access to social attention, access to tangibles, sensory stimulation, or escape from aversive stimuli. In accordance with research regarding technology use and autism (Mazurek et al., 2016; Mazurek & Engelhardt, 2013; Mazurek & Wenstrup, 2013), access to electronic devices pre- and post-bedtime was a common contributing factor toward participant sleep disturbance. In fact, Isaac, Ben, and Finn were the only participants whereby device use was not implicated in their sleep disturbance. Notably however, despite experiencing similar sleep problems, the specific functions underlying sleep-interfering behaviour varied across participants, emphasising the importance of conducting an FBA. The current study adds to the small body of literature demonstrating the effectiveness of sleep interventions informed by FBA and their applicability within natural settings, such as the home (Didden et al., 2002; Friedman &

Luiselli, 2008; Jin et al., 2013; McLay et al., 2017; McLay, France, Blampied et al., 2019; McLay, France, Knight et al., 2019; Moore, 2004; Weiskop et al., 2001; Weiskop et al., 2005).

Importantly, many FBA-informed treatment components in the current study were directed to and implemented by the young person. These components included psychoeducation, imagery, protective item, bibliotherapy, bedtime routine, stimulus control, relaxation, bedtime pass, and reinforcement. Over the past six decades, only a few other studies have actively included young people with ASD in the psychological treatment of their sleep disturbance, despite this being commonplace for typically developing young people. Steps taken to involve young people with ASD in the management of their sleep problem reflects the basic human right for all individuals to be actively included within society, regardless of ability (UN, 2006).

In the current study, numerous modifications were made to facilitate the active involvement of young people with ASD in the therapeutic process. This included tailored communication methods (e.g., use of closed, open, and multiple choice questions, indirect communication [e.g., text, email, letters], providing extended time periods to respond, clarifying participant understanding); use of visual resources (e.g., social stories, checklists, cartoons, scales/thermometers, written resources, VSG); parent attendance during sessions; consistent structure and routine for each session (e.g., outlined agenda at beginning of each session, contacted participant at the same time each day); and utilisation of special interests. Participants demonstrated receptive, expressive, and written communication abilities far below the level demonstrated among their typically developing peers. Accordingly, in line with previous research involving young people with ASD, behavioural treatment components were used more commonly than language-mediated cognitive components (Burkhart et al., 2018; Ho et al., 2015; Walters et al., 2016; Weston et al., 2016). Promisingly, research suggests adolescents find behavioural treatment components of CBT-I more helpful than cognitive components (Blake, Sheeber et al., 2017). Although participants with lower communication abilities required more parent input, as is consistent with existing literature regarding application of CBT to young people with ASD (Burkhart et al., 2018; Ho, Stephenson, & Carter, 2014; Keefer et al., 2018; Kester & Lucyshyn, 2018; Perihan et al., 2019; Walters et al., 2016), young person-implemented treatment components were feasible when appropriate modifications were carried out.

Previous research has demonstrated the effectiveness of parent-implemented behavioural sleep interventions for children with ASD. The findings presented in this thesis suggest a combined parent- and young person-implemented approach, and occasionally a young person-implemented only approach, can successfully improve sleep disturbance. However, the design of the three studies in this thesis precluded direct comparison between parent-implemented and young person-implemented intervention, therefore the contribution of young person-implemented techniques is unknown. Parent input is critical to assessment and in almost all cases was necessary in varying degrees to effectively treat sleep disturbance. Participants too played a vital role in informing intervention as well as acquiring skills to self-manage their sleep problem. Ideally, a shared parent-child management approach to treatment should be utilised, enabling each party to upskill, contributing to treatment maintenance, and likely facilitating generalisation to the management of other behaviour difficulties.

The Treatment Acceptability of FBA-informed Young Person- and Parent-implemented Sleep Interventions

Within empirical research, a strong emphasis is placed on intervention effectiveness, often with minimal consideration of the social validity of such procedures (Callahan et al., 2017; McNeill, 2019). Although there is an increasing evidence base for behavioural sleep interventions, social validity is vital to the application of treatment (Callahan et al., 2017; McNeill, 2019; Snodgrass, Chung, Meadan, & Halle, 2018). Wolf's (1978) seminal paper suggests interventions should be socially important (e.g., improvement to the dependent variable is a valued goal), appropriate (e.g., cost-effective, practical, applicable in natural settings), and effective (e.g., inducing clinical change, Horner et al., 2005). Further, the perception of all consumers and direct participants of an intervention should be considered. This includes both the young person and their caregiver/s (Callahan et al., 2017; Kazdin, 2000; Snodgrass et al., 2018; Wolf, 1978).

Existing research suggests ASD-related behavioural sleep interventions and CBT-I are generally acceptable to caregivers (Carnett et al., 2019; Kirkpatrick et al., 2019; McCrae et al., 2019), however, the young person's perception of treatment acceptability has rarely been assessed. In fact, the current study is the first within the autism and sleep literature to report young people's perspective of treatment acceptability. Overall, based on YPTE scores and post-treatment interviews, participants considered the assessment and treatment

procedures to be moderately to highly acceptable. These findings are concordant with literature that includes typically developing individuals, which also suggests young people consider cognitive and behavioural treatment components for insomnia to be acceptable (Bei et al., 2013; Blake, Sheeber et al., 2017; Bootzin & Stevens, 2005; de Bruin et al., 2014).

According to TARF-R results, parents generally considered treatment to be effective, reasonable, and affordable. During post-treatment interviews parents emphasised the value of young person-implemented treatment components. Primarily, they were perceived to be less invasive and facilitative of autonomous sleep management. This supports recent research which indicated parent-reported acceptability for combined parent-child implemented sleep treatment was high (Loring et al., 2016; McCrae et al., 2019). In contrast, some caregivers within the current research considered parent-implemented treatment (e.g., extinction of tangible items) to be far more problematic. Crucially, social validity may have contributed to intervention effectiveness and maintenance. Parents and young people who were the least supportive of intervention, also admitted inconsistent implementation, potentially contributing to a deterioration in target sleep variables at follow-up. Findings emphasise the appropriateness of a collaborative model, such as the guided participation model (Sanders & Burke, 2014), whereby consumers are actively involved in treatment decisions enabling families to implement interventions which are feasible, practicable, and supported by them.

Some participants considered therapist contact to be boring and/or "annoying" when it prevented engagement in preferred activities, with one participant indicating he would have preferred to receive support from a parent instead. Conversely, 7/12 participants enjoyed being able to talk to and receive support from a person outside the family. Therapist-client alliance is a strong predictor of treatment outcomes though it may be influenced by the core symptoms of ASD, such as poor reciprocal social interaction (e.g., exchange of social niceties), thus making it harder for clinicians to engage young people with ASD in treatment. Further, in the current study, high rates of social anxiety among participants necessitated parent coaching with minimal direct contact from the therapist at times. On the other hand, rigid adherence to rule following meant some participants were more likely to comply with therapist-initiated techniques as opposed to parental advice and therefore therapist involvement was crucial. The current findings indicate the level of therapist and/or parent support offered should be responsive to the individual needs and desires of each family. This

is also consistent with the principles of FBA, which preclude a one-size-fits-all intervention approach.

A key theme evident from TARF-R results and interviews with participants and their parents was ensuring intervention was not too invasive or time intensive. Although FBA results may necessitate rigorously applied and comprehensive interventions, this may not be realistic for many families with children on the autism spectrum. Further, clinicians are unlikely to be able to facilitate long-term interventions, particularly when they are laborious. On average, intervention lasted for 63 nights/9 weeks (range, 28 – 106 nights/ 4 – 15 weeks). Within the literature, the reported duration of behavioural sleep interventions for young people with ASD has ranged from 7.5 to 67 weeks (Carnett et al., 2019), compared with 2 to 8 weeks for typically developing children (Mindell et al., 2006). It is possible that the complexities of everyday life for families with a child with a neurodevelopmental disorder may prolong treatment and delay the acquisition of new behaviours. Despite minimally sufficient interventions being feasible (as illustrated in Study 3), they may not be effective for complex families. Nevertheless, Beresford et al. (2016) found parents in their study favoured high-intensity, individualised interventions which were informed by a comprehensive assessment of their unique situation, as is consistent with an FBA approach. Crucially, the current research showed young person-implemented treatment components can reduce reliance and lessen the need for more restrictive practices.

Interventions can be considered socially valid if the significance of the treatment effects justifies the methods used (Wolf, 1978), as long as such methods are ethical. The current study revealed intervention resulted in both statistical and clinically significant change in socially important dependant variables. This included improvement in sleep disturbance as well as reliable reductions in problem behaviour and anxiety. Further, at post-treatment some participants became able to attend sleepovers outside the home, parents were able to share their bed with each other, as well as receive uninterrupted sleep and more time to themselves in the evenings. Additionally, many families noted there was less stress and conflict during the morning and evening routines. Although Blair's family were in the intervention phase for the longest, they reported it was worthwhile and they would recommend other families receive similar support.

Overall, findings suggest FBA-informed interventions involving both participant and parent input may be both effective and socially valid. Further, the current research provides

new evidence which illustrates the value of young person-implemented treatment components.

Participant and Parent Wellbeing Post-treatment

Very few studies have investigated the impact of sleep interventions on the daytime behaviour and functioning of young people with ASD. Secondary outcomes for participants in the current study consisted of reliable reductions in overall problem behaviour (e.g., attention, thought, and social difficulties) as well as internalising difficulties (e.g., anxiety and depressive symptoms). The reason for these improvements may be that such behaviours were inadvertently targeted via the sleep intervention. For example, learning to sleep independently may have enhanced participant self-efficacy regarding management of anxiety-provoking situations. Equally, application of relaxation skills at bedtime may have facilitated emotional regulation during the day. In accordance with existing literature, the current research provides further evidence of the bidirectional relationship between sleep and mood disorders in young people with ASD (Nadeau et al., 2015). It also supports the idea that internalising problems, such as excessive worry, may play a role in the initiation and maintenance of sleep disturbance in older children and adolescents with ASD (Richdale et al., 2014). Interestingly, externalising behaviours such as aggression, were not reliably different at post-treatment and did not appear to contribute towards sleep problems in many of the participants. Additionally, perhaps unsurprisingly, there was not a reliable change in ASD symptom severity for most participants post-treatment. Given autism is a neurodevelopmental disorder, it would be unlikely to find a significant reduction in symptoms which typically persist throughout the lifetime. It is also possible that the secondary outcome measures used in this thesis were not sufficiently sensitive to detect change in each area.

Despite research revealing strong associations between paediatric sleep disturbance and poor parental wellbeing, few studies have examined the impact of ASD-related sleep interventions on the wellbeing of caregivers. Interestingly, the findings of the current research suggested parents did not necessarily have clinically substantive sleep, mental health, or marital relationship difficulties pre-treatment and therefore, the resolution of sleep problems did not significantly impact their overall wellbeing. Anecdotally however, at post-treatment parents reported reduced conflict at bedtime and during the morning routine, as well as subsequent reductions in associated stress. The relationship between sleep and wellbeing is likely mediated by numerous factors and therefore it is not surprising there was an

inconsistent relationship between improved sleep and wellbeing in the current study. Nevertheless, in some instances, resolving ASD-related paediatric sleep disturbance may have collateral benefits for the wellbeing of the entire whānau/family and therefore can be considered a valuable component of psychological treatment.

Additional Research Findings

Measurement of Sleep. While not an aim of this research, it became apparent that the widely used sleep measures in the current study provided inconsistent and/or contradictory information at times. It is of critical importance that family members and health professionals alike can reliably detect and accurately measure sleep problems. Existing research has largely relied on parent-report data to identify sleep problems, inform treatment, and evaluate treatment success (Moore et al., 2017). A strength of the current research is that sleep was assessed via a range of measures (parent- and self-report sleep diaries and questionnaires, clinical interviews, VSG), enabling assessment of their congruence. Importantly, findings revealed inconsistency between information obtained from these sources, calling into question the reliability and validity of these measures. VSG appeared to provide data additional to parent-report measures, perhaps due to the essentially private nature of sleep-related phenomena in older children and adolescents. Respondent error (e.g., reporting wake time incorrectly, miscalculation of CCs) may have also contributed to the incongruence between parent- and/or self-reported measures compared with VSG. Nevertheless, VSG is not without its faults. The accuracy of video-observed data can be compromised by blurry or obstructed views (e.g., face covered by blanket). Further, video-observed data cannot be obtained if the young person moves location during the night (e.g., transitions to parents' bed; Moore et al., 2017). Very few sleep-related questionnaires have been specifically developed for use by young people with ASD and their parents or standardised on relevant samples (Moore et al., 2017), potentially contributing to the collection of inaccurate information. The findings of the current study illustrate the importance of triangulating information, especially when screening for sleep disturbance, as measures used in isolation may not reliably detect the magnitude of the issue.

Telehealth. Although telehealth delivery was not a focus of the present research, many of the participants resided in different geographical locations throughout NZ, necessitating non face-to-face (indirect) modes of communication (e.g., video call, text messaging, email, mail) during assessment and intervention. Existing research in this area

indicates standard parent-mediated behavioural sleep interventions can be effectively delivered via telehealth, facilitating flexibility and responsiveness (McLay, Sutherland, Machalicek, & Sigafoos, 2020; Stuttard, Clarke, Thomas, & Beresford, 2015). Digital application of CBT-I with typically developing adolescents has also been promising, resulting in treatment effects comparable to face-to-face treatment (Blake et al., 2019; de Bruin et al., 2014; 2015; McLay et al., 2020). No previous research has evaluated digital CBT-I with young people with neurodevelopmental disorders (Blake et al., 2019), however, indirect modes of therapy may be preferable for youth with ASD, preventing social anxiety often generated by face-to-face therapy. Within the current research, use of indirect communication methods meant it was difficult to engage with participants remotely when they were distracted by their environment (e.g., playing video games). Further, during telephone calls the therapist was not necessarily aware of the cause of the distraction and it was difficult to intervene effectively. Technological communication with parents was also challenging when their attention was consumed by household activities (e.g., child behaviour management). Despite these challenges, sufficient data was able to be collected from families who did not reside in the same area as the therapist or have access to the University clinic in order to conceptualise and inform intervention, as well as monitor treatment effects. Although the impact of mode of intervention delivery on treatment outcome was not formally assessed in the current study, all participants experienced improved sleep regardless. Overall, the current research provided preliminary evidence to suggest FBA-informed sleep interventions can be delivered via telehealth, reducing travel burden, promoting treatment in home settings, and enabling families in rural settings to access support from experienced clinicians/specialist services (McLay et al., 2020).

Limitations

It is paramount that all individuals on the autism spectrum have access to effective psychological therapy for sleep disturbance, however, the generalisability of the current research has yet to be determined. There are a number of methodological limitations which compromise the external validity of the findings. Firstly, the present research consisted of a small number of participants, therefore, there has been a limited number of replications of the treatment and its effects. Further, these replications involved the same research programme. To establish treatment efficacy on the basis of single-case research, Chambless and Hollon's (1998) guidelines state treatment effects must be replicated multiple times by independent

researchers. Secondly, the lack of female participants inhibits generalisability across genders; although, the ratio of male to female participants in the current research is consistent with diagnosis rates globally (Loomes et al., 2017). Thirdly, as cultural differences (e.g., family sleep practices, general child behaviour management strategies) may mediate sleep treatment outcomes, the research findings may not be applicable to many families in NZ due to its cultural diversity.

Another key limitation of the current research is the inconsistent and missing data, limiting the reliability and validity of the results. Parent-reported sleep diary data was collected for all participants, however information regarding target variables was not necessarily reported each night. Further, although parent-report diary data was triangulated with multiple sources (e.g., parent and self-report questionnaires, clinical interviews, VSG), additional information was not always available. For example, when participants did not consent to VSG, the reliability of parent-report could not be established. Additionally, due to various factors (e.g., invalid measure for certain age ranges, questionnaire availability, missing responses), psychometric measures were not completed consistently, for example, not all parents completed the CSHQ. Use of standard questionnaires across participants would have enabled better comparison, however, valid self-report questionnaires for a wide age range are not readily available. Lastly, given the lack of correspondence between sleep diary data and video-observed data, IOA for video-observed data could have helpfully been conducted to ensure reliability.

As parent- and young person-implemented treatment components were applied simultaneously, it was not possible to isolate the treatment effects of either approach. Most studies relating to the involvement of young people with disabilities in research provide the authors' opinions about the impact of this inclusive practice (Bailey et al., 2015). Future quantitative research could gather objective data regarding the specific impact of child involvement, although this is likely to prove difficult given certain components are not easily classified as parent- or young person-implemented (e.g., white noise). Further, the developmental stage and presence of a neurodevelopmental disorder, are likely to necessitate parent input of some kind.

Future Research

Given the age range and communicative abilities of the participants in the present research, it is not known whether the assessment and intervention approaches would be appropriate for young children and young adults on the spectrum, or those who are non-verbal. Existing research suggests FBA-informed sleep interventions are efficacious for young children with ASD, consequently future research could evaluate the use of procedures in the current study with older adolescents and young adults. This is another neglected area of research. Very few studies have examined sleep interventions with older age groups, despite research suggesting sleep disturbance is a lifelong condition without treatment (Goldman et al., 2017). Such research is paramount to promoting the independent functioning of individuals with ASD in adulthood.

Notably, the intellectual functioning of participants was not formally assessed in the current research. Therefore, its contribution to treatment outcomes is unknown. Future research could conduct formal cognitive assessments (e.g., Wechsler Intelligence Scale for Children, Fifth Edition, Wechsler, 2014) with participants to determine whether cognitive ability per se predicts the effectiveness of young person-implemented intervention. Additionally, future research could explore further methods which support the active inclusion of all young people with ASD, regardless of cognitive ability. Equally, additional communication methods (e.g., picture exchange system; Frost & Bondy, 1994) could be trialled to assess applicability to young people with reduced verbal ability. Researchers suggest cognitive behavioural approaches to treatment could be applied to individuals on the spectrum who are non-verbal and/or have lower intellectual abilities if a functional communication system is used (Attwood & Scarpa, 2013). Preliminary research in this area is promising (Pardini et al., 2012).

The current study's findings illustrate triangulation of data is necessary to ensure an accurate understanding of sleep problems; however, increasing demand and stretched resources within the mental health sector limits clinician capacity to conduct comprehensive assessments. Consequently, it may not be feasible for practitioners to carry out thorough FBAs as in the current research, involving the collection and interpretation of a range of information sources. Nevertheless, although conducting FBA can be time-consuming, it may be critical to the development of effective interventions. Future research is needed to identify the simplest and most accurate measures of sleep-related dependent variables. Currently, parent- and self-report sleep diaries are likely the most easily accessible, inexpensive, and

non-time-consuming continuous measures of sleep, although, they may not be sufficiently accurate. Evaluation of training procedures to enhance the validity of parent- and self-report via sleep diaries may be warranted.

As yet, few psychometric measures for sleep have been normed on individuals with ASD, thus compromising their utility with this population. Future research could focus on the development of psychometrically sound instruments for the assessment of sleep and related variables in young people with ASD. Consumer-rated social validity is critical to the evaluation of efficacious treatment. Nevertheless, scant research has considered young peoples' perception of sleep interventions. It is recommended that future studies assess the social validity of parent and young person-implemented behavioural sleep interventions based on the perspective of both parent and child. This would assist in the clarification of whether a shared management approach to sleep disturbance is considered more acceptable than traditional parent-implemented interventions.

Conclusion

Despite the preceding limitations, the current research makes a number of important contributions to the existing literature. Firstly, it illustrates the effectiveness and maintenance of FBA-informed sleep interventions for older children and adolescents with ASD. Secondly, it demonstrates verbal young people with ASD can take an active role within the assessment and therapeutic process of their sleep problem, reducing reliance on parents for data collection and treatment implementation. Thirdly, this shared parent-child management approach to sleep disturbance is reportedly acceptable to caregivers and young people alike. Finally, improved sleep was associated with significant reductions in internalising behaviour as well as attention, thought, and social problems. Overall, the findings indicate FBA-informed, young person-implemented treatment components may be an appropriate first-step to address ASD-related sleep disturbance in older children and adolescents and may have positive implications for their mood and behaviour. Further investigation is necessary to evaluate the efficacy of less restrictive and minimally sufficient FBA-informed approaches, although initial findings are promising. Future studies with larger sample sizes, more replications, robust designs, and participants diverse in culture, ASD symptom severity, communicative ability, and cognitive functioning are necessary to generalise the findings of the present research.

Whāia te iti kahurangi, ki te tuoho koe me he maunga teitei

Pursue that which is precious, and do not be deterred by anything less than a lofty mountain

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Appendix A: Ethics Approval

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

HUMAN ETHICS COMMITTEE
Secretary, Rebecca Robinson
Telephone: +64 03 369 4588, Extn 94588
Email: human-ethics@canterbury.ac.nz

Ref: HEC 2018/47

23 July 2018

Dr Laurie McLay
Health Sciences
UNIVERSITY OF CANTERBURY

Dear Laurie

The Human Ethics Committee advises that your research proposal "An Investigation into the Effectiveness of Treatments for Sleep Disturbance in Children With Autism" has been considered and approved.

Please note that this approval is subject to the incorporation of the amendments you have provided in your email of 16th July 2018.

Best wishes for your project.

Yours sincerely

pp. R. Robinson

Professor Jane Maidment
Chair
University of Canterbury Human Ethics Committee

Appendix B: Flyer

Does your child have autism or features of autism, and sleep problems?

You may be eligible to receive treatment through the nation-wide Autism and Sleep Study, being conducted through the University of Canterbury

- Our research is investigating (a) the effectiveness of treatments for sleep disturbance for children with autism (b) the impact of successful treatment on parent and child well-being (c) the effectiveness of treatments for sleep-interfering repetitive behaviours
- Our research team is led by Dr Laurie McLay and Associate Professor Karyn France, and consists of Child and Family Psychology and PhD students
- Sleep treatments can include a range of strategies, including both non-traditional approaches (such as white noise), behavioural interventions and cognitive behavioural therapy.
- Treatment options will be outlined for you, and the final decision will be yours. If the approach is unsuccessful, alternative treatment options will be offered.
- The research will be conducted in the family home or at a University clinic, and will be implemented by parents, with support and guidance through skype and phone calls from the researchers

If you or somebody you know might be interested in participating in this study and you would like further information, please contact:

Dr Laurie McLay
School of Health Sciences, University of Canterbury
Phone: (03) 369 3522
Email: laurie.mclay@canterbury.ac.nz

Jenna van Deurs (PhD Candidate & Registered Intern Psychologist)
School of Health Sciences, University of Canterbury
Phone: (03) 366 7001 and ask for extension 3696
Email:
jenna.vandeurs@pg.canterbury.ac.nz

Appendix C: Parent Information Sheet

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

An investigation into the effectiveness of treatments for sleep disturbance in children with autism or features of autism

Information for Parents/Caregivers

This research has been assessed and approved by the University of Canterbury Human Ethics Committee (HEC 2014/150).

Dear Parent/ Caregiver,

We are a group of researchers at the University of Canterbury. Dr Laurie McLay is a lecturer in the School of Health Sciences at the University of Canterbury. Laurie has many years experience in working with children and young people with autism and their families. Associate Professor Karyn France has lectured here for many years, has conducted research into the treatment of paediatric sleep disturbance and is a registered clinical psychologist with considerable clinical experience in this area. Professor Neville Blampied has a similar history of teaching and research. Jolene Hunter and Jenna van Deurs are PhD candidates, and Jemma Vivian is working on her thesis as part of this project.

We would like you to consider allowing your child with autism or features of autism to participate in this research study. The primary purpose of this study is to investigate the effectiveness of treatments for sleep disturbance in children with autism. Treatment can include a range of strategies, including non-traditional approaches (such as white noise),behaviour interventions and cognitive behavioural therapy. These approaches have been designed to minimise stress as much as possible for the parents and children using them. We are also interested in parents' experiences in using the treatments and any changes to their lives, or their child's lives, which result.

If you agree to allow your child to be a part of this study, we will meet with you to discuss your child's sleep behaviour and find out more about him/her and your family. This initial meeting will last for approximately 1-1 ½ hours. We will then ask you to complete sleep diaries in which you will record further information about your child's sleep patterns. Sleep diaries will be recorded each day throughout all phases of the study as this will allow us to monitor the effectiveness of the treatment approach. The sleep diaries will take you up to five minutes to complete each night. You will also be asked to complete commonly used questionnaires in order to obtain information about your child's sleep behaviour and the effects of treatment. It will take approximately 15 minutes to complete each questionnaire. When we have established an understanding of your child's sleep behaviour, we will work with you to develop sleep-related goals for your child. This will involve a second treatment planning meeting which will last 1-1 ½ hours.

To help us gather further information about your child's sleep patterns we will bring a video camera to your home for some nights over the course of the programme, which is capable of recording all night sleep. In addition we will ask you, if possible, to use an actigraph with your child. This watch-like device records the movements associated with sleep and can be worn on the wrist or ankle, or secured into a pocket on your child's pyjamas. These methods will allow us to measure sleep behaviour at times when an adult is not present. We will demonstrate and explain how to use each of these methods for gathering information.

As a part of this study we would also like to investigate the experiences of families in implementing treatments for sleep disturbance, those treatments that they consider to be most acceptable, and the impact of successful treatment of sleep problems on parent and child wellbeing and quality of life. In order to do this we will ask you to complete some questionnaires about you and your child's well-being and behaviour at the commencement and conclusion of treatment. We will also ask your perspective on the treatment that was provided. We will do this either during visits to your home or in a clinic at the University of Canterbury.

When information about your child's sleep behaviour has been gathered, treatment will commence. You will be offered a choice of treatment options. If you are dissatisfied with the treatment approach or the degree of progress that is being made then you will be offered a choice of another treatment option. We will provide you with all of the necessary information about each treatment approach and we will maintain regular contact with you during treatment. It is anticipated that your involvement in the study will occur over the course of a few months, but will depend on the rate of your child's progress as well as your satisfaction with the progress.

Your child will be assigned a code name to ensure anonymity and anything that you or your child says or does will be kept confidential. The results of the study may be submitted for publication to national or international journals and may also be presented at conferences.

If you want to withdraw from the project before completion, you can do this at any time without penalty or repercussions.

Should you require any additional information about the study or if you would like to access the study findings you are able to do so at any stage. The data which is produced from the research will be kept in a locked cabinet at the University of Canterbury for a minimum of ten years.

If you agree for your child to take part in the research, please sign the consent form that is attached.

If you have any complaints you may contact the Chair of the University of Canterbury Ethics Committee. The contact details are given below.

If you have any questions about this project please feel free to contact;

Dr Laurie McLay: Phone 64 (3) 364-2987 ext. 7176, Email laurie.mclay@canterbury.ac.nz
or Jenna van Deurs: Phone 64 (3) 366-7001 state ext. 3696; or 021 080 74246, Email jenna.vandeurs@pg.canterbury.ac.nz

Appendix D: Parent Consent Form

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

CONSENT FORM FOR PARENTS/ CAREGIVERS

This research has been assessed and approved by the University of Canterbury, Human Ethics Committee.

I wish to participate in the project, *"*An investigation into the efficacy of treatments for sleep disturbance in children with autism*"*

I have read and understood the information that was given to me about this study. I understand what will be required of myself and my child/the child in my care during this project

I understand that the investigators do not foresee any potential risks to me or my child as a result of participating in this study

I understand that all information about my family will be treated as confidential unless there is concern about anyone's safety. In this case my clinician will need to speak to someone else to ensure the safety risk is removed. No findings that could identify me or my child will be published

I understand that the findings of this study may be published in a research journal or at a conference and that the anonymity of my child and I will be maintained

I understand that participation in this project is voluntary and that I can withdraw my child or he/she can withdraw from the project at any time without repercussions. I can also withdraw any data that has been collected at any time prior to the publication of that data

I understand that all research data that is collected will be securely stored at the University of Canterbury for a minimum of ten years

I understand that I am able to request a copy of the results of this research, should I wish to do so, and that these results will be provided for me

I allow video-taping of my child's sleep behaviour to be completed by the researcher and understand that this videotape will be used for data gathering purposes only. I also understand that I have the right to request that video footage is destroyed at any stage.

I consent to others, listed below, being involved in the implementation of the intervention

Name: _____

Date: _____

Signature: _____

Others I consent to implementing intervention:

Name:_____

Name:_____

Name:_____

Please return this form to Jenna van Deurs.

Appendix E: Parent Audiovisual Recording Consent Form

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

An Investigation into the Efficacy of Treatments for Sleep Disturbance in Children with Autism

AUDIOVISUAL RECORDING CONSENT FORM

You have been given this form because the researchers have asked your permission to take audiovisual recordings of your child's sleep behavior.

Please read the statements below, which explain the purpose of audiovisual recording and how your privacy will be protected:

- The purpose of recording is to gather data for the research project
- Audiovisual recording will only be done with your knowledge and consent
- You can withdraw your consent to audiovisual recording at any time, without having to provide a reason for changing your mind
- The audiovisual file will only be seen by the researchers
- The audiovisual recording will be securely stored at the University of Canterbury for a minimum of ten years

I hereby consent to audiovisual recordings being made on the above conditions.

Signed: _____

Date: _____

Appendix F: Child Information Sheet

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

An investigation into the efficacy of treatments for sleep disturbance in children with autism

Children's Information Sheet

Hello. My name is Jenna van Deurs and I am a PhD student at the University of Canterbury. I am doing a project about how to help children to sleep better and I would like for you to help me with this.

I am going to be talking to you and your parent/s about ways to help you to sleep better. This means that I might be Skyping you, coming to your house, or your parent/s will be coming to see me at the University.

There will be a video camera in your bedroom sometimes. This will help me to understand what you do when you are awake and asleep. Only your parents and other people working on this project will be able to see this video. We may ask you to wear an actigraph. An actigraph is worn on your wrist like a watch and it tells us when you are asleep and when you are awake.

If you do not want to be a part of this project, you can tell me or your parents and you won't need to be a part of it anymore.

If you have any questions you can ask me or your parents whenever you like.

Now we need to decide if you would like to do this. If you do want to be a part of my project then you can say "yes". If you do not want to be a part of this project then you can say "no" and no one will mind.

If you say yes, you or one of your parents can sign the form for you.

This research has received ethical approval from the University of Canterbury Human Ethics Committee, Private Bag 4800, Christchurch; email human-ethics@canterbury.ac.nz

Appendix G: Child Consent Form

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

"An investigation into the efficacy of treatments for sleep disturbance in children with autism"

Children's Consent Form

My name is _____.

Jenna has told me about the work that she is going to be doing with me and my parent/s.

Jenna told me that she is going to be working with me and my parent/s to help me to learn to sleep better.

I know that if I want to stop at any time or if I do not want to be a part of this project anymore that will be fine. I can tell Jenna or my parents.

I was told that my parents/caregiver may sign this form for me and I think that is OK.

Child's name: _____

Date: _____

Signature: _____

If this form is signed on behalf of your child please acknowledge, by signing this form, that your child was verbally informed of the investigation and what it will involve and that they were unable to provide verbal or written consent that they would like to be a part of this research.

Parent/caregiver: _____

Date: _____

Signature: _____

Please return this form to Jenna van Deurs.

This research has received ethical approval from the University of Canterbury Human Ethics Committee, Private Bag 4800, Christchurch; email human-ethics@canterbury.ac.nz

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

An investigation into the efficacy of treatments for sleep disturbance in children with autism

Young Person Information Sheet

Hello. My name is Jenna van Deurs and I am a PhD student at the University of Canterbury. I am doing a project about how to help young people sleep better and I would like for you to help me with this.

I am going to be talking to you and your parent/s about ways to help you sleep better. This means I might be Skyping, phoning or texting you, coming to your house, or your parent/s will be coming to see me at the University.

I will ask you to complete some questionnaires so I can find out more about your sleep and the impact it may be having on other areas of your life.

There may be a video camera in your bedroom sometimes. This will help me to understand what you do when you are awake and asleep. Only your parents and other people working on this project will be able to see this video. We may ask you to wear an actigraph. An actigraph is worn on your wrist like a watch and it tells us when you are asleep and when you are awake.

If you do not want to be a part of this project, you can tell me or your parents at any time and you won't need to be a part of it anymore.

If you have any questions you can ask me or your parents whenever you like.

If you would like to be a part of my project then you can sign the attached form. If you do not want to be a part of this project then you can say "no" and no one will mind.

This research has received ethical approval from the University of Canterbury Human Ethics Committee, Private Bag 4800, Christchurch; email human-ethics@canterbury.ac.nz

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

An investigation into the efficacy of treatments for sleep disturbance in children with autism

Young Person Consent Form

My name is _____.

Jenna has told me about the work she is going to be doing with me and my parent/s.

Jenna told me she is going to be working with me and my parent/s to help me learn to sleep better.

I know if I want to stop at any time or if I do not want to be a part of this project anymore that is fine. I can tell Jenna or my parents.

Date: _____

Young Person's Signature: _____

Please return this form to Jenna van Deurs.

This research has received ethical approval from the University of Canterbury Human Ethics Committee, Private Bag 4800, Christchurch; email human-ethics@canterbury.ac.nz

Appendix J: Young Person Audiovisual Recording Consent Form

UNIVERSITY OF CANTERBURY
Te Whare Wānanga o Waitaha
CHRISTCHURCH NEW ZEALAND

An Investigation into the Efficacy of Treatments for Sleep Disturbance in Children with Autism

VIDEO/ ACTIGRAPH RECORDING CONSENT FORM

We would like to make video/actigraph recordings of your sleep to help gain information for our project.

Video recordings are often used to record sleep because of the detailed information they give. We use a special video which works in the dark.

An actigraph is a watch-like device which measures movement. It gives us information about your sleep pattern, like how much time each night you spend in different kinds of sleep.

Video/actigraph recordings will help us better understand your sleep difficulties and show us any changes in your sleep over time.

We will only record you with your permission and will always let you know when we are recording.

You can ask us to stop recording at any time for any reason.

Only people involved in the project can view the recordings.

If you agree to video/actigraph recordings being made please sign below:

Signed:

Date:

Appendix K: Example Parent Clinical Interview Content

Introduction

- Explanation of role and Autism Sleep Study team
- Confidentiality
- Agenda

Child Information

- Ethnicity/culture
- Interests/strengths
- Typical daily routine
- Schooling
- Medication

Presenting Problem

- Typical sleep routine
- Bedtime resistance
- Sleep environment
- Sleep-interfering behaviour (antecedents and consequences)
- Night wakings
- Early wakings
- Frequency/intensity of sleep problem
- Exceptions to problem
- What makes it better/worse

History of Sleep Problem

- Onset, what happening in life at this time
- Attempts to address problem and outcome
- How has problem changed over time

Parent Relationship

- Attributions regarding sleep problem
- Responses to child's challenging behaviour
- Agreement across parents regarding attributions and behaviour management
- Ability to work as a team
- Current relationship strength

Developmental History

- Life prior to pregnancy
- Pregnancy
- Birth
- Post-birth adjustment
- Milestones
- Physical health
- Social behaviour
- Academic skills
- Major events

Parent Mental Health

- Impact of sleep problems on parent sleep, mood etc
- Other factors/stressors contributing to wellbeing
- Management strategies
- Support

Set Sleep Goals

Appendix L: Example Young Person Clinical Interview

Introduction

- Explanation of role and Autism Sleep Study team
- Purpose and agenda
- Confidentiality

Interests, hobbies, likes and dislikes

Family

- People in household
- Relationships

School

- Location
- Teacher
- Classmates
- Subjects

Sleep

- Bedtime routine
- Bedtime resistance
- Sleep position
- Sleep environment (e.g., conditions needed to fall asleep)
- Sleep-interfering behaviour (antecedents and consequences)
- Perceived sleep problems and history
- Emotion knowledge and understanding of cognitions
- Cognitions at bedtime and throughout the night
- Nightmare content
- Sleep quality
- Energy levels in morning and throughout the day

Sleep goals

- 3 wishes/spells to change sleep
- Motivation to attain goals
- Confidence will attain goals

Appendix M: Parent Sleep Diary

Sleep Diary

Child's Name:

	Date:	Monday:	Tuesday:	Wednesday:	Thursday:	Friday:	Saturday:	Sunday:
Daytime sleep	Setting (where fell asleep)							
	Time asleep							
	Time awake							
Night-time sleep	Setting (where fell asleep)							
	Time put to bed							
	Frequency of Curtain calls*							
	Curtain calls after put to bed (Describe each)							
	Your responses to each curtain call (Describe each)							
	Best estimate of time asleep							

		Monday	Tuesday	Wednesday	Thursday	Friday	Saturday	Sunday
1st Night time awakening	Time & Duration of awakening	_____ mins	_____mins	_____ mins	_____mins	_____ mins	_____ mins	_____ mins
	Behaviour while awake (Describe)							
	Your responses (Describe)							
2nd Night time awakening	Time & Duration of awakening	_____ mins	_____mins	_____ mins	_____mins	_____ mins	_____ mins	_____ mins
	Behaviour while awake (Describe)							
	Your responses (Describe)							
Time awake in the morning								

*Curtain calls: Any behaviour such as leaving the bed (or bedroom) or calling parents into the room, between the time of being put to bed and falling asleep

Notes:

Appendix N: Example Young Person Sleep Diary

Eve's Sleep Diary

	Monday	Tuesday	Wednesday	Thursday	Friday	Saturday	Sunday
Bedtime							
Number of times I called out or left my bedroom							
Time I fell asleep							
Number of times I woke up							
Length of time I was awake							
Time I woke in the morning							

Appendix O: Example Social Story

Peter's Social Story

Page Number	Sentence Content	Corresponding Picture
1	Peter's Social Story	Peter asleep in bed
2	Sleep is really important. Getting enough sleep makes me healthier, stronger and happier.	Cartoon pictures of a smiley face, muscles flexing, and Joy (character from Pixar's Inside Out)
3	Staying awake during the day helps me feel sleepy at bedtime. If I stay awake all day, I will receive a treat a night.	Peter sitting on the couch awake
4	Before I get ready for bed, I can use my phone, laptop, and iPad as much as I like downstairs.	Peter using his laptop on the couch
5	Because my phone, iPad, and laptop are exciting, noisy, and full of light, they make my brain alert. When my brain is alert it is hard to fall asleep.	Cartoon of child sitting up in bed looking frustrated
6	When I put my devices away at night, I fall asleep faster and sleep through the night.	Peter asleep in bed
7	At 10.15 pm I put my laptop, iPhone, and iPad into the Finished Box because it is time for bed.	Finished Box with laptop, iPhone, and iPad inside
8	I put my devices in the Finished Box straight away and get a chocolate. If I have stayed awake all day, I get another chocolate AND if I use kind words and keep my hands to myself when I put my devices in the Finished Box, I get another chocolate.	M&Ms
9	My phone, iPad, and laptop are in a safe place overnight and I can play with them in the morning at 7am. I can sleep without my phone, laptop, and iPad.	Closed Finished Box

Page Number	Sentence Content	Corresponding Picture
10	After I brush my teeth, I get into bed. To fall asleep, I lie in bed with my eyes closed. I can breathe slowly and use the exercises in my Relax Book to help me feel relaxed and sleepy.	Peter asleep in bed
11	If I wake during the night, I can make myself relax by doing slow breathing and the exercises in my Relax Book. I feel relaxed, so I go back to sleep.	Relax Book
12	When my alarm clock goes off at 7am it is time to get up. At 7am I can go downstairs and can use my phone, laptop and iPad again!	Alarm clock showing 7:00am and Peter using his laptop on the couch
13	When I go straight to sleep and sleep through the night without my iPhone, iPad and laptop my family are so proud of me! They might ring other people to tell them what a great job I am doing.	Mother and sister smiling and making thumbs up gesture

Appendix P: Example Sleep Checklist

Ben's Sleep Checklist

☐ Have a drink of water, or place a water bottle beside my bed

☐ Have a snack if needed

☐ Use the toilet

☐ Check my door is open enough

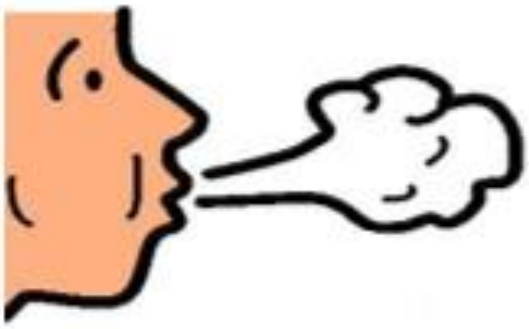
☐ 8:30pm bed

Appendix Q: Example Incorporation of Young Person's Interests

Excerpt from Peter's Relax Book (special interest: Pixar)

Breathe out like a steam train

1



2

3

4

5

Excerpt from Seth's Relax Book (special interest: 1920s steam trains)

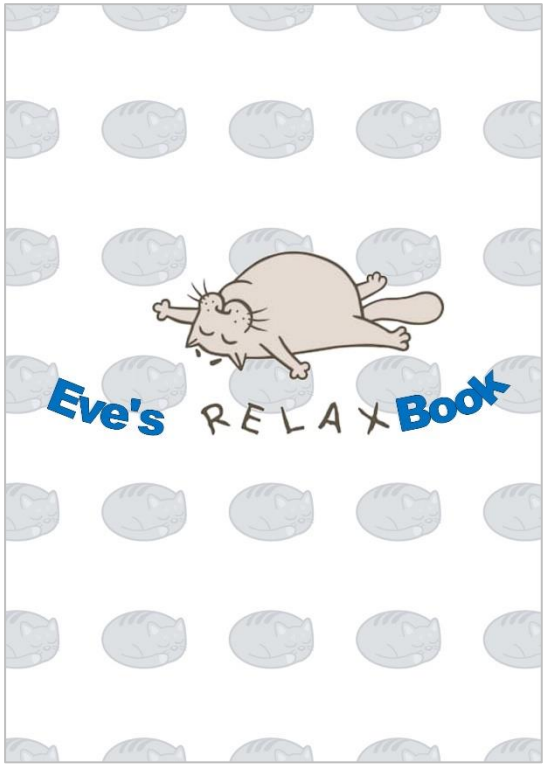
Appendix R: Young Person Treatment Evaluation Form

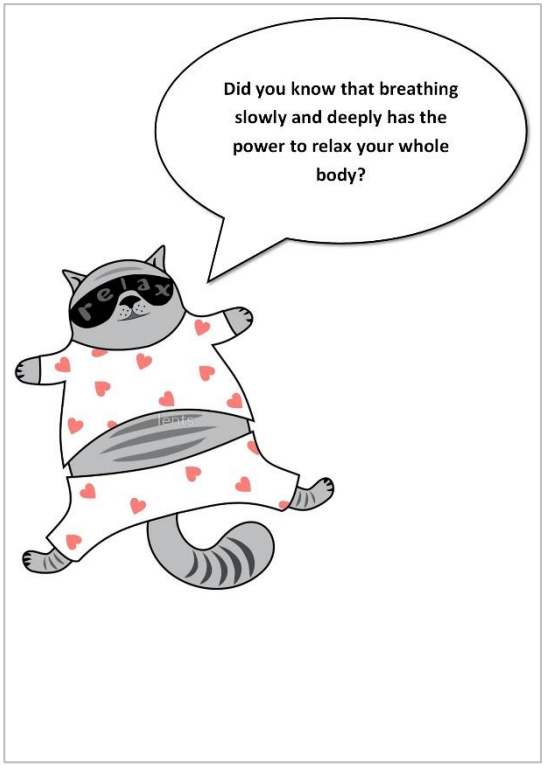
Child/ Young Person Treatment Evaluation

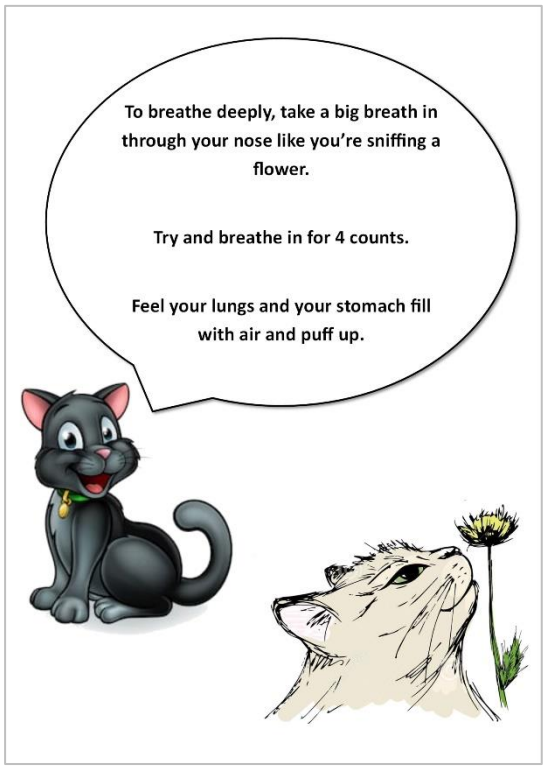
Instructions: Please circle the correct answer to the questions below

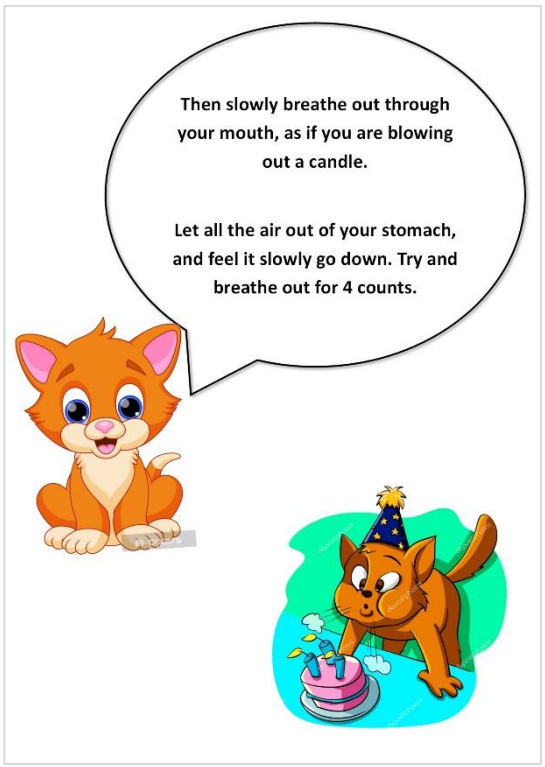
1) How helpful was the sleep treatment?	**Not at all helpful** 	**OK** 	**Very helpful** 
2) How much has your sleep improved?	**Not at all** 	**OK** 	**Very much** 
3) How much did you enjoy the sleep treatment?	**Not at all** 	**OK** 	**Very much** 
4) How much time was needed to do the sleep treatment?	**Too much time** 	**Neither too much or little time** 	**Little time** 
5) How fair was the sleep treatment?	**Unfair** 	**OK** 	**Very fair** 
6) How did you find the sleep treatment overall?	**Bad** 	**OK** 	**Good** 

Appendix S: Eve's Relax Book[5]









[5] Not to scale

Keep slowly breathing in and out.

Breathe in 1, 2, 3, 4

Breathe out 1, 2, 3, 4

Focus on your breathing

Keep going until you feel calm and relaxed.

Now that you're breathing calmly, let's relax your muscles.

Stretch your legs out in front of you as far as they can go, curl your toes and hold for a few seconds.

Release and feel your legs relax and sink into the bed.

Imagine a dog is running along not watching where he is going. He is about to step on your stomach. Quick tighten your stomach muscles and make them hard.

Phew the dog has gone, you can relax your stomach.

Stretch your arms out in front of you, raise them up high over your head. Stretch as far as you can go.

Then drop them to your sides and feel them relax into the bed.

Imagine a fly has landed on your nose and you need to wrinkle your nose and scrunch up your face to get it off. Scrunch up your face.

Loosen the muscles in your face. Feel your head relaxed on the pillows.

Think about how your body feels now you have let all the tightness in your muscles go.

You feel floppy, loose and relaxed and ready for a snooze.

Appendix T: Example Parent Post-treatment Interview

Post-treatment Interview

1. How did you find the intervention overall, and the process?

2. What is it that you (both) did, that you feel made a difference?

3. How do you make sense of the improvement?

4. Did the child's progress/improvement have an impact on you personally, if so- how? What impact did it have on the child/ the rest of your family?

5. On a scale of 1-5 with 5 as the worst the sleep problems could possibly be, what level of impact do you see the sleep problems as having currently (1 being 'no problem' and 5 being 'very much a problem- big impact')

6. Did you notice any other changes in your child's behaviour in response to intervention?

7. Any suggestions for how our process could have been improved?

8. Any other comments you would like to make:

Appendix U: Example Young Person Post-treatment Interview

Post-treatment Interview

1. When we first met your sleep was…. What, if anything, has changed about your sleep since we started working together?

2. How is your sleep now?

3. What did we do to help your sleep?

4. Tell me what you thought about X treatment strategy:

5. What was most helpful? Least helpful?

6. What did you like most? What did you like least? ‘

7. What could have been done differently?

8. If I was working with another person who was X years old like you and it was taking them a long time to fall asleep and they were waking in the night how could they make their sleep better?

9. Other comments:

Appendix V: van Deurs, J. R., McLay, L. K., France, K. G., Blampied, N. M., Lang, R. B., & Hunter, J. E. (2019). Behavioral sleep intervention for adolescents with autism spectrum disorder: a Pilot study. Advances in Neurodevelopmental Disorders, 3, 397–410. doi:10.1007/s41252-019-00123-z

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ORIGINAL PAPER

Behavioral Sleep Intervention for Adolescents with Autism Spectrum Disorder: a Pilot Study

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Abstract

Objectives Sleep disturbances are a significant problem for individuals with autism spectrum disorder (ASD) across the lifespan; however, there is a paucity of research examining effective sleep interventions for adolescents with ASD. Although research has demonstrated individuals with ASD can be meaningfully engaged in their own intervention process, such engagement has not appeared in previous studies targeting sleep in adolescents with ASD.

Methods This study investigated the feasibility of including pre-adolescents and adolescents (ages 9 to 14 years) with ASD as active intervention agents within comprehensive, individualized treatments for sleep problems. Participants had a range of intellectual functioning but all produced spoken language. Outcomes were evaluated using single-case designs.

Results Data suggest intervention was effective in eliminating sleep disturbance for all participants. Improvements were maintained during 18- to 24-month follow-up. All three participants and their parents indicated a high degree of treatment satisfaction.

Conclusions Findings illustrate the feasibility and potential benefit of including adolescents with ASD in the process of developing and implementing individualized behavioral interventions for sleep problems.

Keywords Autism spectrum disorder · Sleep assessment · Sleep treatment · Adolescent · Functional behavior assessment

Difficulties initiating and maintaining sleep as well as other topographies of sleep disturbance are a common clinical problem for individuals with ASD and their families. In a study of 1518 children with autism spectrum disorder (ASD), Malow et al. (2016) found that 71% had clinically significant sleep problems, a much higher rate than in samples of typically developing individuals (e.g., Elrod et al. 2016). Although the exact cause of sleep problems in people with ASD likely varies across individuals, previous research suggests their sleep is impacted by a complex interaction between physiological (e.g., dysregulated melatonin), environmental, and behavioral (e.g., inadvertent reinforcement of sleep-interfering behavior) variables (Richdale and Schreck 2009). A range of sleep problems have been identified in people with ASD including sleep onset delay, reduced total sleep time, reduced sleep efficiency, and daytime fatigue (Baker et al. 2013). These issues appear to be particularly problematic during adolescent years (Baker et al. 2013; Goldman et al. 2012). This is consistent with findings involving typically developing adolescents wherein physiological vulnerabilities (e.g., changes in circadian phases), environmental factors (e.g., increased internet use, responsibilities at school and work), schedule changes (e.g., early school start times), and increased autonomy (e.g., freedom to choose their own bedtime and bedtime routine) appear to increase risk for sleep disturbance (Loring et al. 2016). These risk factors can be exacerbated among individuals with ASD whereby deficits in executive functioning may inhibit the organization and regulation of sleep-conducive bedtime behavior (e.g., following a consistent bedtime routine, limiting caffeine consumption, restricting screen use at night; Quist et al. 2015). In addition, preliminary research suggests adolescents and young adults with ASD are more likely to have dysregulated levels of melatonin compared with typically developing controls (Tordjman et al. 2012).

Clinically significant sleep problems are detrimental to the overall wellbeing of individuals and their families. Specifically, insufficient sleep has been linked to increased

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severity of autism symptomatology, challenging behavior, and poor psychological wellbeing, as well as compromising parental sleep, quality of life, and marital relationships (Cortesi et al. 2010; Herrmann 2016). Without effective intervention, sleep problems experienced by adolescents with ASD may persist over time, compromising future functioning across home, school, and work settings.

Behavioral intervention and parent-education programs have been effective in treating sleep disturbance in children with ASD (Cuomo et al. 2017). Increasingly, these interventions are informed by Functional Behavior Assessment (FBA), whereby an FBA is used to identify the variables influencing sleep problems and inform selection of individualized, multi-component, function-based treatments (e.g., Jin et al. 2013; McLay et al. 2019). Although there is evidence to support the use of function-based treatments for sleep problems in young children with ASD, there is little research on the utility of behavioral sleep interventions for adolescents with ASD. Further, FBA-informed sleep intervention has not been investigated with adolescents with ASD. Instead, pharmacological approaches, in particular melatonin, have been the most thoroughly researched treatment (Cuomo et al. 2017).

Most existing sleep-intervention research with typically developing adolescents involves the young person as the primary change agent. Specifically, involving young people in the sleep intervention increases their knowledge of sleep-conducive behavior and teaches them skills to resolve their own sleep disturbance (Schlarb et al. 2011). A review of the autism and sleep literature revealed that only one study has actively included adolescents with ASD in the therapeutic process. Loring et al. (2016) provided two sleep-education sessions to 18 adolescents (11 to 18 years old) with high-functioning autism (HFA, IQ > 70) and their parents. Session one targeted sleep hygiene including bedtime routine and arranging the sleep environment, and session two taught relaxation and distraction techniques to facilitate sleep onset. Subsequent application of these practices by the adolescents, with parental support, resulted in significant improvements in sleep onset and efficiency. In other studies, young people with ASD have been engaged in the sleep-intervention process through social stories and visual schedules (e.g., Delemere and Dounavi 2018; Moore 2004). However, in those cases, parents were the primary intervention agent.

When modifications are made to standard therapies to facilitate the engagement of adolescents with ASD, it is important to assess the social validity and acceptability of the procedures and include input from the adolescents (Callahan et al. 2010). Unfortunately, most social validity data have been collected via parent report, even though parents did not directly experience the targeted sleep disturbance or treatment. Further, reliability between parent-reported sleep diaries and objective sleep measures (e.g., videosomnography) has predominantly been assessed with younger pre-adolescent children. Parents may be less likely to detect an adolescent's covert sleep-interfering behavior (e.g., electronic device use), and the validity of parent-reported sleep outcomes should not be assumed.

A number of questions remain about the engagement of adolescents with ASD and sleep disturbance in their own treatment. First, the practicalities of including adolescents with ASD, across a range of intellectual functioning, in the assessment and treatment process are not well established. Second, the long-term effectiveness of behavioral sleep interventions for adolescents with ASD is understudied; specifically, there appears to be no study with follow-up beyond 12 months (Durand 2002; Weiskop et al. 2001). Third, little is known regarding adolescent perspectives and social validity of behavioral sleep interventions, which would seem particularly important when interventions include components delivered directly to the participant. Finally, although parent-reported sleep diaries are considered reliable among preschool and school-aged children (Hodge et al. 2012), the reliability of parent-reported sleep in adolescents with ASD warrants consideration. The present study evaluates (a) outcomes of individualized behavioral sleep interventions involving input from adolescents with ASD; (b) long-term maintenance of effects; (c) social validity of treatment components implemented with adolescents for sleep disturbance; and (d) the reliability and validity of parent-reported sleep diaries for adolescents with ASD.

Method

Participants

Participants ranged in age from 9 to 14 years old; although one participant was pre-adolescent, participants will be referred to as adolescents hereafter. Participants were referred to the research study by their parents or by professionals delivering services to individuals with ASD and their families. Each participant met the following inclusion criteria: (a) formal diagnosis of ASD or Asperger's syndrome, as verified by a psychiatrist, registered psychologist, or pediatrician, and supported by results of the Gilliam Autism Rating Scale, Third Edition (GARS-3, Gilliam 2013); (b) parent-reported difficulty initiating and maintaining sleep, supported by systematic in-home measurement; (c) no medical condition that directly interfered with sleep; and (d) sufficient receptive and expressive communication skills to engage in treatment. The communication criterion was assessed through clinical judgment coupled with item responses on the Communication domain of the Vineland Adaptive Behavior Scales Second Edition, Caregiver Rating Form (VABS-II; Sparrow et al. 2005). For example, the Vineland items considered included, "Follows instructions with one action and one object" and "Says at least

Table 1 A summary of participant characteristics at commencement of intervention

Characteristics	Niko	Peter	Eric
Age (Y-M)	9–7	14–6	11–6
Gender	Male	Male	Male
Diagnosis	Asperger's syndrome	ASD	ASD
VABS-II Receptive and expressive language age equivalent (Y-M)	2–5 4–11	2–10 3–11	5–6 12–3
Educational environment	Mainstream school (teacher aide support)	Specialist school	Mainstream school
GARS-3	118 Very likely	108 Very likely	106 Very likely
CBCL (6–18 years)	Clinical	Clinical	Clinical
Medication	–	Melatonin (3 mg) Risperidone (0.25–0.5 mg) Fluoxetine (20 mg)	–

50 recognizable words". Participant characteristics are summarized in Table 1 (pseudonyms have been used in place of real names to protect participant privacy).

Procedures

Experimental Design A single-case design, incorporating baseline [A], intervention [B], and short- and long-term follow-up phases, was used to evaluate treatment effects. Additional intervention phases were indicated by a phase-change line and alphabetization (C, D, or E). An AB design was applied to Niko and Eric, and an ABCDE design to Peter.

Setting Participants were located throughout New Zealand. Clinical interviews and treatment planning discussions with families were conducted in a university-based clinic or at the participant's home if they were unable to travel to the clinic. The VABS-II and GARS-3 were administered to caregivers over the phone prior to the clinical interview. Other pre-treatment psychometric assessments were given to the parent at the clinical interview to complete and return to the researcher. Post-treatment questionnaires were sent to families upon conclusion of the intervention phase. Treatment was implemented within the participant's home by adolescents and their parents with the support of the first author. During treatment, communication with participants and families was conducted in person or via Skype, telephone, and email contact, depending on geographical location and participant preference.

Functional Behavioral Assessment A combination of the Sleep Assessment Treatment Tool (SATT; Hanley 2005), Questions About Behavioral Function (QABF; Matson and Vollmer 1995), sleep diaries, and analysis of video footage was used to conduct the FBA. The SATT is an open-ended interview tool designed to identify factors contributing to children's sleep disturbance; it was used to guide questions in the clinical interview. The QABF, a brief functional assessment checklist used to establish the function of a target behavior, was completed by parents following the clinical interview. Information gathered about the history and type of sleep problems, sleep hygiene practices, antecedent and consequence variables maintaining the sleep problem, and its possible function was synthesized in an FBA-informed case conceptualization (Blampied 2013).

Baseline Baseline commenced following completion of the FBA. Baseline length was staggered such that Peter, Niko, and Eric completed 3, 5, and 8 weeks of baseline recording respectively. Baseline length was determined by random assignment, though on occasion this was extended due to participant readiness to commence intervention (i.e., to ensure that the conclusion of baseline was commensurate with the beginning of treatment, baseline was occasionally extended). During baseline, families were asked to continue with their existing sleep practices (e.g., bedtime routine, electronic device use).

Intervention Individualized, FBA-based, multi-component interventions commenced upon conclusion of baseline. The chosen treatment was discussed with parents and participants using the guided participation model to ensure a shared understanding of the issues and intervention (Sanders and Burke 2014). Treatment included components implemented with both parent and participant. Each treatment component was selected to address hypothesized factors underlying participant sleep disturbance, facilitate treatment compliance, and support the maintenance of helpful sleep habits. Table 2 includes a summary of each participant's sleep problem, FBA data, and subsequent intervention components.

During the intervention phase, researchers communicated daily with parents. Daily (Niko) and weekly (Peter and Eric)

Table 2 Problem behavior, factors precipitating and/or maintaining behavior, hypothesized function, and parent and adolescent treatment components for all three participants

	Niko		Peter		Eric
	Frequent and prolonged NWs	Frequent EWs	Delayed SOL	Frequent EWs	Delayed SOL
Factors thought to be precipitating and/or maintaining behavior	Daytime sleeps; lack of physiological sleep pressure; electronic device use; adolescent-reported discomfort in bed; warm and comfortable sleep-interfering environment	Daytime sleeps; lack of physiological sleep pressure; electronic device use; adolescent-reported discomfort in bed; warm and comfortable sleep-interfering environment	Daytime sleeps; lack of physiological sleep pressure; inappropriate sleep dependencies (electronic device, bright light); lack of discriminative stimuli for sleep; electronic device use; exposure to bright nightlight	Daytime sleeps; lack of physiological sleep pressure; inappropriate sleep dependencies (electronic device, bright light); lack of discriminative stimuli for sleep; electronic device use; exposure to bright nightlight	Lack of physiological sleep pressure; exposure to bright light and stimulating content on electronic devices; access to food and drink; parent responses to CC's; intrusive internal stimuli; hyperarousal
Hypothesized function	Tangible Escape	Tangible Escape	Tangible	Tangible	Tangible Social attention Escape (from intrusive internal stimuli)
Parent treatment components	Sleep hygiene; bedtime fading and sleep restriction (elimination of naps); modified extinction (removal of devices at night and scheduled device use); comfortable sleep setting; positive reinforcement for successive approximations towards goal	Sleep hygiene; bedtime fading and sleep restriction (elimination of naps); modified extinction (removal of devices at night and scheduled device use); comfortable sleep setting; positive reinforcement for successive approximations towards goal; Gro-Clock	Sleep hygiene; graduated extinction (removal of devices at night); bedtime fading and sleep restriction (elimination of naps); Finished Box; sleep item; replacement of nightlight; consistent sleep cues; positive reinforcement	Sleep hygiene; graduated extinction (removal of devices at night); bedtime fading and sleep restriction (elimination of naps); Finished Box; sleep item; replacement of nightlight; consistent sleep cues; positive reinforcement	Restricted access to devices after dinner; modified extinction (minimal engagement post-bedtime); bedtime fading and sleep restriction (set sleep and wake times); positive reinforcement
Adolescent treatment components	Psychoeducation; social story; relaxation techniques	Psychoeducation; social story; relaxation techniques	Psychoeducation; social story; relaxation techniques	Psychoeducation; social story; relaxation techniques	Psychoeducation; relaxing bedtime routine; sleep checklist; relaxation techniques; visualization

contact was also maintained with adolescent participants. During regularly scheduled contact, researchers provided participants with feedback regarding treatment implementation, treatment fidelity was monitored, and praise and encouragement was given. The intervention continued until the participant's sleep disturbance had been significantly reduced or eliminated, and this pattern had been evident consistently across a 10- to 14-day period. The intervention phase lasted for 48, 94, and 84 nights respectively for Niko, Peter, and Eric. FBA results and individualized treatments are detailed below (additional details regarding interventions are available from the first author).

The FBA revealed numerous antecedent and consequence variables appeared to be interfering with participants' sleep. These included lack of physiological sleep pressure due to inconsistent sleep/wake times and daytime sleep; stimulating activities pre-bedtime; presence of intrusive internal stimuli (e.g., reports of distressing cognitions); inappropriate sleep dependencies (e.g., electronic devices); lack of discriminative stimuli for bedtime (e.g., inconsistent bedtime routines); sleep environment discomfort; and exposure to light. Reinforcement contingencies for sleep-interfering behavior came in the form of parental attention; access to electronic devices and other preferred items (e.g., food and drink); and purported escape from intrusive internal stimuli (reduced distress).

Intervention components implemented with all three participants included discussions about key sleep-facilitative behaviors (e.g., closing eyes), the impact of sleep disturbance on areas of importance to them (e.g., ability to play video games well), and connection between their behavior and sleep

disturbance. All three also received instruction in relaxation training (e.g., progressive muscle relaxation [PMR], deep breathing) which has been suggested to reduce physiological arousal and support independent sleep onset (Stewart and Gordon 2014). All parents received psychoeducation regarding the relationship between existing operant and respondent conditioning processes and their child's sleep disturbance, sleep hygiene (Jan et al. 2008), modified extinction (Kuhn and Weidinger 2000), and positive reinforcement strategies.

Varying topographies and functions of sleep problems necessitated the use of additional individualized intervention components. Individualized components implemented included (a) social stories to depict new targeted sleep routines, sleep-conducive behavior, and reinforcement contingencies (Gray and Garand 1993); (b) sleep checklists (a visual schedule of the bedtime routine); (c) visualization techniques to redirect sleep-interfering cognitions; and (d) Gro-Clocks (to provide a discriminative stimulus for sleep/wake times). Verbal instruction, social stories, modeling, visual aids (e.g., picture cues), parent presence, and participant interests were integrated to facilitate participants' comprehension and engagement with therapeutic resources. Individualized parent-mediated interventions included (a) bedtime fading and sleep restriction (Vriend et al. 2011) to increase physiological sleep pressure and create a motivating operation for sleep; (b) appropriate sleep dependencies (provision of sleep-conducive stimuli that were accessible throughout the night e.g., a soft toy; Jin et al. 2013); (c) clear discriminative stimuli for bed preparation and sleep onset (e.g., consistent statements about bedtime and sleep); (d) scheduled access to putative reinforcers (Jin et al. 2013); (e) graduated extinction (Vriend et al. 2011); and (f) positive reinforcement for successive approximation towards desired sleep behavior.

Niko Niko's primary sleep problems included frequent and prolonged night wakings (NWs) as well as early wakings (EWs, i.e., any rise time before 6:00am where sleep was not reinitiated). FBA results suggested Niko's sleep disturbance was maintained by access to tangible reinforcement (electronic device use) and escape from the perceived discomfort and cold of his bed to a heated lounge. Lack of physiological sleep pressure due to daytime sleep was also suggested to be contributing to Niko's sleep difficulties.

The following treatment components were implemented simultaneously: sleep hygiene; psychoeducation; a social story; a comfortable sleep environment; bedtime fading and sleep restriction; relaxation techniques; modified extinction (restricted access to electronic devices and lounge heater); Gro-Clock; and positive reinforcement for successive approximation towards desired sleep behavior (e.g., remaining in bed until successively later from 5:00am). The preceding techniques functioned to increase sleep pressure, reduce reinforcement for sleep-interfering behavior, and promote engagement in sleep-conducive behavior. Niko showed a reluctance to stop using electronic devices during the night, despite psychoeducation. As access to electronic devices was hypothesized to be the primary reinforcer of sleep-interfering behavior, restricted access was imperative. Niko was taught relaxation techniques, as a replacement behavior for leaving his bedroom when he woke, to facilitate reinitiation of sleep. Niko agreed a Gro-Clock was necessary to signal appropriate sleep/wake times as he was unable to read the time, enabling him to earn rewards. Immediate reinforcement for improvements in sleep outcomes was provided to strengthen Niko's participation in therapy, as his parent indicated he would become unmotivated and non-compliant rapidly without immediate reinforcement for progress. Reinforcement options were collaboratively agreed on by Niko and his parent.

Peter Peter's primary sleep problems included delayed sleep-onset latency (SOL) and EWs thought to be reinforced by access to preferred items (e.g., electronic devices). Additional precipitating and maintaining factors included lack of physiological sleep pressure, inappropriate sleep dependencies (e.g., electronic device use), and lack of discriminative stimuli for bedtime. Intervention phase one consisted of teaching Peter relaxation strategies and providing a social story. These were implemented first to provide a rationale for and prepare Peter for later changes, provide him with skills to manage anxiety, and facilitate sleep onset. These components also reduced parent anxiety regarding Peter's capacity to cope with change and thereby enhanced treatment fidelity.

Phase two of intervention included consistent implementation of appropriate sleep dependencies (e.g., soft toy); discriminative stimuli for sleep (e.g., switching off the bedroom light at bedtime); and gradual extinction of mobile phone use, achieved by scheduled access to all of his devices until 15 min prior to bedtime at which point he was asked to place his mobile phone in a visually enticing "Finished Box," and was reinforced for compliance (immediately provided an edible treat, e.g., chocolate coin). Peter chose a sleep item (e.g., soft toy) to take to bed with him, as an appropriate sleep dependency, and his parents issued a consistent sleep statement and turned off his bedroom light. Peter's bright nightlight was replaced with a dim one to facilitate melatonin secretion and reduce the visibility of preferred items, while continuing to provide a source of comfort. Bedtime fading and elimination of daytime sleep was an additional component implemented to ensure sufficient physiological sleep pressure.

Intervention phases three and four included the above components plus the gradual extinction of Peter's iPad and then laptop respectively. Reinforcement for compliance was faded as Peter learnt to independently put his devices away and go to bed on time. Phase five consisted of fading Peter's bedtime earlier in 15-min increments to an age appropriate time.

Eric Eric's primary sleep problems included frequent curtain calls (CCs, i.e., bids for parental attention) and a delayed SOL. His FBA revealed his sleep disturbance was maintained by both positive and negative reinforcement contingencies, including access to tangibles (electronic devices and food), social attention, and by sleep-interfering cognitions (e.g., "What if something happens to Mom when I'm asleep?"). Antecedent variables implicated in Eric's sleep disturbance included lack of physiological sleep pressure, hyperarousal, and exposure to bright light and stimulating electronic device content prior to bed. Exposure to bright light from electronic devices immediately before bedtime may have interfered with Eric's natural melatonin secretion. Furthermore, exposure to device content was suggested to interfere with Eric's ability to reach a relaxed state.

Intervention included simultaneous implementation of psychoeducation (instruction regarding the importance of sleep, and the impact of sleep-interfering and sleep-conducive behavior); a sleep checklist; restricted access to electronic devices prior to bedtime; modified extinction (minimal parent response to CCs); bedtime fading and sleep restriction; and relaxation and visualization (taught to picture a pleasant/peaceful scene during sleep onset). Following consultation with the researcher, Eric agreed to stop using electronic devices after dinner and further enforcement of restrictions was not necessary. Eric's sleep checklist supported his tendency for rule-following and bedtime routine compliance. Collaboration regarding checklist items provided Eric some control over his bedtime routine and was intended to increase his motivation to adhere to intervention. Giving him a relaxing bedtime routine, relaxation instruction, and sleep restriction functioned to reduce the hypothesized association between bed and hyperarousal. Eric's mother was also instructed to minimize interactions post-bedtime and Eric was taught relaxation skills to facilitate independent management of sleep-interfering cognitions and reduce reinforcement for sleep-interfering behavior.

Short- and Long-term Follow-up Short- and long-term follow-up data were collected for 1 week using sleep diaries at 3 to 5 weeks and 12 to 13 weeks post-treatment. An 18- to 24-month follow-up phone interview was conducted with parents at which time they were also asked to begin a 7-day sleep diary. However, only Eric's family completed the extended follow-up diary.

Measures

Clinical Interviews Separate clinical interviews were conducted with each family and participant prior to commencing baseline. Interviews were supplemented with visual aids to support communication. Information on past and present sleep disturbance, participant developmental history, family context, and possible environmental (e.g., bedtime routine) and mental health factors (e.g., anxiety) that could be interfering with sleep were discussed.

Parent-Reported Sleep Diaries Parents recorded data in daily sleep diaries during each phase of the study. Diaries were used to record (a) frequency and duration of daytime sleep; (b) duration of SOL; (c) frequency of CCs; (d) frequency and duration of NWs; and (e) time of morning waking. The latter was used to calculate discrepancy between actual and goal wake time and identify incidents of EW. Participants' sleep setting, behavior during CCs and NWs, as well as parents' responses to this behavior were also noted. Sleep diaries were returned to the research team on a weekly basis.

Videosomnography Swann Advanced-Series DVR4-1200, nighttime, infrared video cameras were used to record participant's sleep and to permit the coding of interobserver agreement (IOA) data. Information obtained from video included (a) topographies of awake behavior post-bedtime (e.g., vocalizations, stereotypy, play); (b) topographies of sleep behavior (e.g., sleep position, eye movement, limb movement); (c) duration of SOL; (d) frequency of CCs; (e) frequency and duration of NWs; and (f) time of morning waking. The following operational definitions were used to code video (a) asleep, lying down with minimal non-discrete movement, and no indication of wakefulness; and (b) awake, the presence of any sleep-interfering behavior, eyes open, or frequent physical movement (Jin et al. 2013). Recording began when the participant went to bed and ended when they awoke to begin the day. Video footage was downloaded to an external hard drive and distributed regularly to the research team to enable objective monitoring of participant progress.

Child Sleep Habits Questionnaire (Owens et al. 2000) The Child Sleep Habits Questionnaire (CSHQ) was completed during assessment and post-treatment to evaluate change in parent-reported sleep disturbance. The CSHQ was completed by Niko and Eric's parents. Peter's parents did not complete the CSHQ as Peter was not within the measure's validated age range. The CSHQ is a parent-report questionnaire consisting of 45 items relating to children's sleep patterns, scores > 41 are indicative of clinically significant sleep disturbance (Owens et al. 2000).

Child Behavior Checklist for Ages 6–18 (Achenbach 2001) The Child Behavior Checklist (CBCL) is a 113-item parent report measure of internalizing (e.g., withdrawn) and externalizing (e.g., aggressive) behavior. The CBCL was completed by parents during assessment and used to indicate the extent of additional behavioral difficulties experienced by participants.

Gilliam Autism Rating Scale The GARS-3 was used to corroborate ASD diagnoses and indicate symptom severity. The GARS-3 is a 56-item informant rating scale of autism symptomatology (Gilliam 2013). It is designed to assess the likelihood a person has ASD and the severity of their behavior in accordance with the Diagnostic and Statistical Manual of Mental Disorders (DSM-5). Items are summed to provide a total ASD Index score. Higher scores indicate a high likelihood and increased severity of ASD.

Treatment Acceptability The Treatment Acceptability Rating Form-Revised (TARF-R; Reimers et al. 1992) was administered post-treatment to assess parents' perceptions of overall treatment acceptability. The TARF-R consists of 17 items which examine ratings of treatment acceptability and three items assessing problem severity and participants' understanding of the intervention approach. Ratings on six subscales (Effectiveness; Reasonableness; Willingness; Cost; Negative side-effects; Disruption/time) are summed to provide a total treatment acceptability score. In addition, parents and their child were interviewed separately to assess the social validity of treatment and to provide qualitative information regarding treatment effects. Information pertaining to sleep (e.g., fatigue, sleep quality), secondary outcomes (e.g., mood), preferred and non-preferred assessment and treatment components, knowledge regarding healthy sleep habits, and suggestions for improvement were gathered. The format of the participant interview included open questions, closed questions with multiple choice options, and visual aids (e.g., photos of treatment components) to facilitate communication.

Interobserver Agreement (See Table 3) Video footage was coded by a researcher blind to parent sleep diary recordings. Agreement between parent report and direct observation data extracted from video was then calculated. Sleep phenomena which parents could not be expected to detect (e.g., covert awakenings in which the adolescent remained quiet in their bed) were omitted from IOA calculations. Measures of duration (e.g., SOL), sleep, and wake times were considered in agreement if parent and video were ± 15 min. Percent agreement for each behavior was calculated using the equation [Agreement/(Agreement + Disagreement)] × 100%. IOA data were collected for 22%, 46%, and 5% of nights across baseline and treatment phases for Niko, Peter, and Eric respectively. Incomplete or lack of sleep diary entries on nights when video data was available, inhibited calculation of IOA on more nights. Limited IOA data were collected for Eric as he withdrew consent for video recording on night 107 due to feeling it was an invasion of his privacy. Niko's mean IOA was 86% for duration of NWs and 100% for duration of EWs. Peter's mean IOA was 93% (range, 85–97%) for SOL and 98% (range, 86–100%) for duration of EWs. Eric's mean IOA was 95% (range, 91–100%) for CCs and 100% for SOL.

Treatment Fidelity (See Table 4) A checklist based on a task analysis of each measurable parent-mediated treatment component (e.g., consistent bedtime) was created for all families. Parent treatment fidelity was assessed by comparing the first author's daily contact notes, video footage, and sleep diaries with the protocol outlined in the treatment checklist. Parent treatment fidelity was calculated for 90% or more of intervention nights across all participants, using the formula (Completed tasks/Total tasks) × 100%. An aggregate score was then calculated for each participant. Treatment fidelity scores were 93%, 98%, and 71% for Niko, Peter, and Eric respectively (mean = 87%). Niko, Peter, and Eric's parents followed every component of the treatment plan (i.e., reached 100% treatment fidelity) on 72%, 84%, and 26% of nights respectively.

Treatment fidelity for each adolescent participant was assessed by examining participant report and video for evidence of treatment adherence. The imperceptible nature of some of the components (e.g., visualization) and unreliable reporting inhibited direct measurement of treatment fidelity; however, there was indirect evidence of intervention compliance by adolescents with ASD. Niko demonstrated mastery of relaxation skills during intervention sessions and was able to identify when to utilize such strategies. However, he was rarely observed to apply these within the sleep context. There was little video evidence of Niko or Peter completing relaxation exercises. After Niko's access to electronic devices was restricted via password protection, he complied with the treatment plan and remained in bed, stating he closed his eyes to reinitiate sleep upon waking as opposed to utilizing relaxation

Table 3 Interobserver agreement (IOA) between sleep diaries and videosomnography across target behaviors

	Niko		Peter		Eric	
Target behavior	Baseline	Intervention	Baseline	Intervention	Baseline	Intervention
CCs	_	_	_	_	91%	100%
SOL	_	_	85%	97%	100%	100%
Duration NWs	_	86%	100%	100%	_	_
Duration EWs	_	100%	86%	100%	_	_

Lack of baseline sleep diary data for Niko inhibited calculation of IOA in this phase

Table 4 Parent treatment fidelity

	Niko	Peter	Eric
Treatment fidelity	93%	98%	71%

strategies. Initial video footage of Eric revealed he completed PMR frequently prior to sleep onset. Eric reported using his sleep checklist and deep breathing strategies every night, although he noted that he did not always stick consistently to his bedtime routine during school holidays.

Data Analyses

Visual Analyses Visual analysis was used to assess the effectiveness of FBA-informed interventions and additional therapies. Level, variability, and trend in the data were evaluated across study phases (Kazdin 2001).

Effect Size Estimate Percentage below the median (PBM) is the percentage of intervention data points below the baseline median (calculated thus because the behavior is decreasing; Parker et al. 2011). For example, if the baseline median was 100 and 25/25, intervention data points were below 100 (PBM = 100%), whereby if 50% of the data points were below 100 (PBM = 50%). Visual analysis was supplemented by calculating PBM to estimate effect sizes: < 70% represents ineffective treatment; 70 to 90% moderate effectiveness; and > 90% high effectiveness (Ma 2009). The reliable change index (RCI) was used to ascertain whether differences in pre- and post-CSHQ scores reflected true significant change as opposed to measurement error (Jacobson and Truax 1991), clinical change occurred when pre-CSHQ scores reduced from the clinical range to the normal range.

Results

Treatment results for each participant are presented individually in a case study format. Sleep diary data for each participant are presented in Figs. 1, 2, and 3.

Data Quality

Video footage was recorded on 20–88% of nights across all participants throughout baseline and treatment phases. Parents recorded sleep diary data for all dependent variables across baseline and treatment on 37–46% of nights (i.e., although

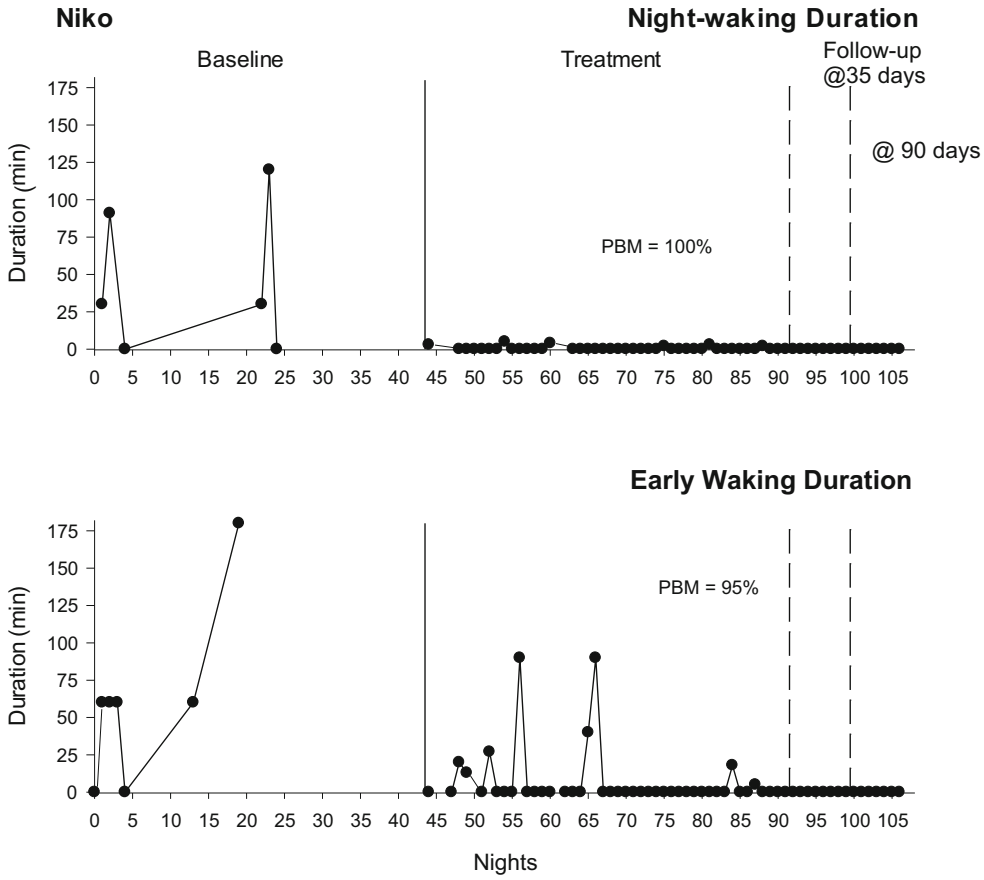

Fig. 1 Sleep outcomes for Niko: duration of NWs and EWs across baseline, intervention, and follow-up phases

parents filled out sleep diaries regularly, they did not collect information on each variable each night).

Niko's baseline sleep diary data is scarce as his parent had difficulty recording diary data during that phase, this prevented calculation of IOA in baseline. There are no sleep diary data for Peter from nights 80 to 102; Peter's parents recorded video only on these nights. Eric's SOL sleep diary data is scarce from night 36 to 85 as school holidays resulted in variability in sleep settings (e.g., tent, friend's house).

Niko

Niko displayed high variability in the duration of NWs (0 to 120 min) and EWs (0 to 180 min) in baseline (Fig. 1). There was an immediate reduction in the level and variability of both sleep variables, with PBM scores (NW = 100%, EW = 95%) demonstrating a large treatment effect. NWs and EWs were eliminated by the end of treatment and these effects were maintained at both short- and long-term follow-up.

At the 24-month follow-up, Niko and his parent reported that the elimination of NWs and EWs had been maintained. Access to devices was still restricted during the night, and Niko used a watch as opposed to a Gro-Clock to signal an appropriate rise time. Additional treatment techniques were no longer required.

Peter

Peter's SOL was highly variable during baseline (range = 5 to 195 min; see Fig. 2). There was an immediate reduction in the level of SOL upon implementation of relaxation strategies. The level and variability reduced further from intervention phases two to four. A large treatment effect was observed within intervention phase four (PBM = 100%). SOL treatment effects were maintained at follow-up. The duration of Peter's EWs was highly variable during baseline (range 0 to 180 min). There was an immediate reduction in the level and variability of the duration of EWs at treatment onset and they were eliminated during treatment, with this result maintained at short- and long-term follow-up.

At the 18-month follow-up, parents indicated that Peter was not experiencing sleep disturbance, SOL was 15 min, and he was not experiencing NWs or EWs. Peter's family still used the Finished Box and restricted his sleep during the day. The social story, sleep item, and reward system were no longer required.

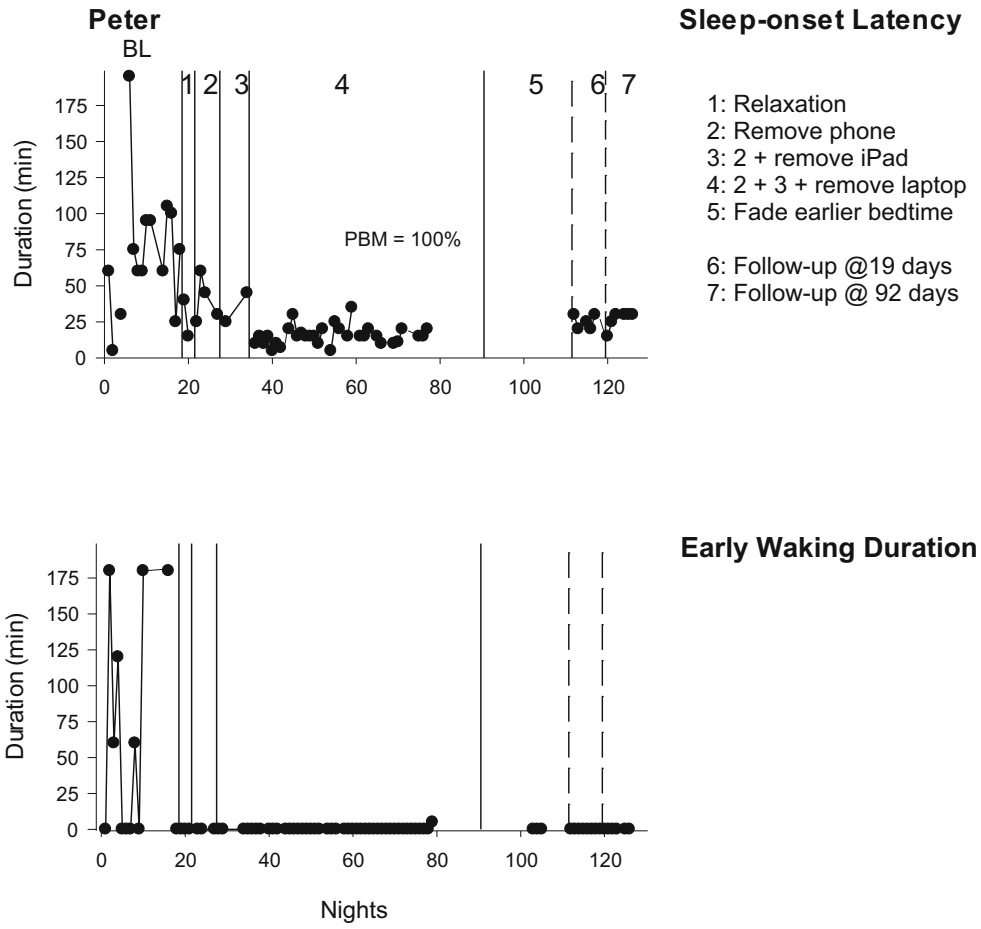

Fig. 2 Sleep outcomes for Peter: SOL and duration of EWs across baseline, intervention, and follow-up phases

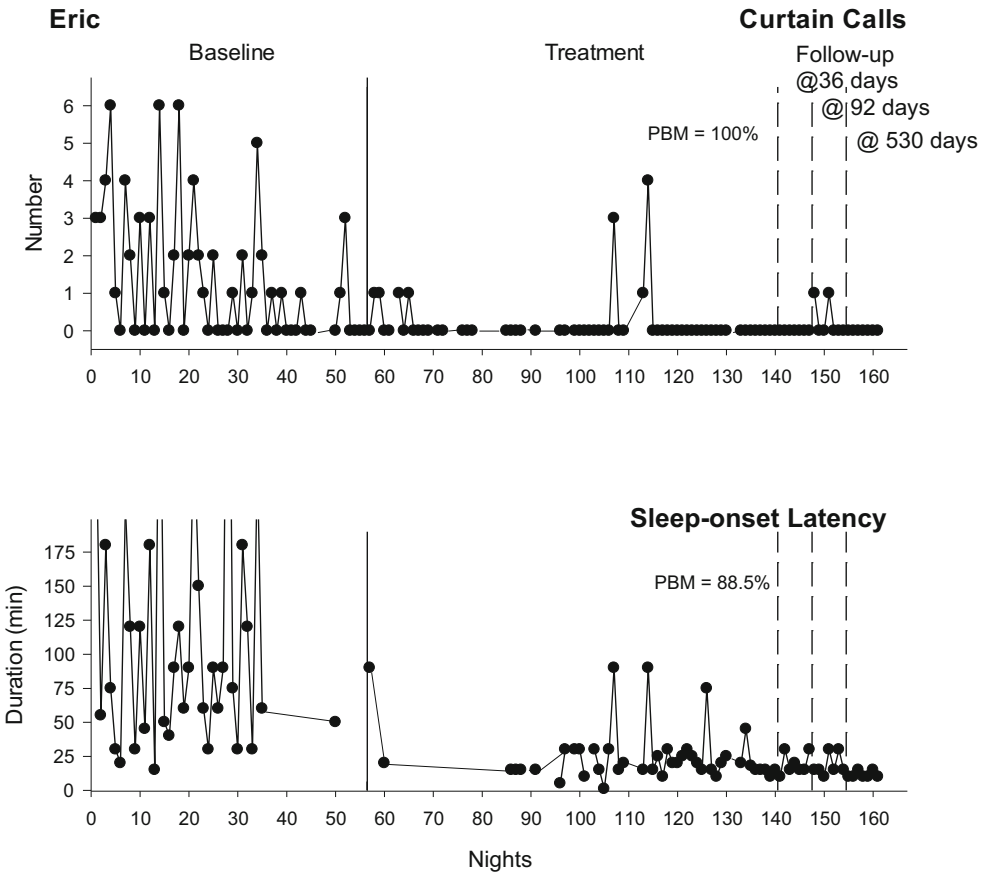

Fig. 3 Sleep outcomes for Eric: CCs and SOL across baseline, intervention, and follow-up phases

Eric

In baseline, Eric displayed a high frequency of CCs and significant variability (range = 0 to 6; see Fig. 3). There was an immediate reduction in the level and variability of CCs with intervention. Treatment had a moderate effect on CCs (PBM = 88.5%). The high frequency of Eric's CCs on nights 107 and 114 occurred during his transition to a new school. SOL was prolonged and highly variable during baseline (range = 15 to 450 min) but reduced significantly during intervention (PBM = 100%), and this was maintained at short- and long-term follow-up.

At 18-month follow-up, Eric's parent reported Eric was not experiencing sleep problems and no longer engaged in bedtime resistance. Eric's mother said his SOL was 10 to 15 min in duration. The family reported continued use of delayed bedtime, restricted access to devices, and use of deep breathing.

Table 5 Pre- and post-treatment CSHQ scores

	Niko	Eric
Pre-treatment CSHQ	52	58
Post-treatment CSHQ	44*	38**

*Significant change; **Clinical change

CSHQ Scores

Both Niko and Eric experienced a reliable change in their CSHQ scores (see Table 5). For Eric, this change was clinically significant, with his post-treatment score falling below the clinical cut-off for sleep disturbance.

Social Validity

Participant reports suggest FBA-informed sleep interventions and components implemented with adolescents were acceptable to participants and their parents. Planned use of tangible and social rewards was well regarded. Participants varied in their rating of relaxation. Eric reported finding deep breathing helpful. He attributed his success to this and noted that engaging in deep breathing at bedtime helped prevent his mind from "buzzing" as he focused on counting his inhalations and exhalations. Niko and Eric reported PMR was ineffective and Peter was ambivalent. Peter was the only participant to indicate he did not like the elimination of electronic device use. Some participants commented on the intrusive nature of the video camera and the discomfort and time-consuming nature of having to talk to a researcher daily. Niko and Eric indicated they had experienced improvements in their sleep, including

Table 6 Post-treatment TARF-R scores

Scale	Niko	Peter		Eric	Maximum Score
	Father	Mother	Father	Mother	
Effectiveness	21	21	21	21	21
Reasonableness	21	19	18	20	21
Willingness	21	18	16	17	21
Cost	14	10	14	14	14
Negative side-effects	21	15	15	15	21
Disruption/time	15	18	16	18	21
Problem severity*	10	11	11	2	14
Understanding of treatment*	7	7	7	7	7
Total acceptability	113	98	94	105	119

*Not included in total acceptability score

reduced SOL and daytime fatigue, as well as ability to reinitiate sleep upon waking.

During post-treatment interviews, all parents reported that the intervention had successfully reduced their child's sleep disturbance and perceived daytime fatigue. Importantly, parents felt their child had developed the skills to manage their sleep independently by implementing "the tools in his strategy bag" (e.g., deep breathing) and adhering to sleep-facilitative behavior (e.g., remaining in bed). Peter's parents reported that, while visual aids were important for his comprehension, they also facilitated parental structure and routine. Eric's treatment, which consisted predominantly of intervention components delivered directly to him, was described by his parent as "non-invasive."

Treatment Acceptability Rating Form-Revised TARF-R scores have a possible range of 17 to 119; higher scores indicate higher acceptability. Parent ratings ranged from 94 to 113 (see Table 6). Each parent's rating yielded the maximum score on the Effectiveness subscale. Overall, parents rated the interventions to be highly acceptable, effective, and easy to understand, taking little time to implement, at no financial cost.

Discussion

The purpose of this study was to evaluate the effects of individualized behavioral sleep interventions involving input from adolescents with ASD alongside parent-mediated treatment with a focus on maintenance and social validity. Further, the validity of parent-report sleep diaries was also assessed. Overall, results of this pilot study suggest that comprehensive, individualized interventions including both adolescent- and parent-implemented treatment components can reduce sleep disturbance. Parents reported that improvements in sleep were maintained at 18 and 24 months post-intervention. However, caution is warranted in interpreting these maintenance data because of reliance on parent report and lack of direct observation. Overall, the treatment components were rated as acceptable (socially valid) by adolescents with ASD and their parents. Specifically, all participants reported that they enjoyed the reinforcement systems and two reported that the adolescent-implemented treatment components (e.g., deep breathing) were beneficial.

Successful application of intervention components delivered directly to the adolescent appeared to have been mediated in part, by each participant's functioning and their use of individualized treatment strategies (e.g., relaxation). Sleep problems resolved to the greatest extent for Eric who had the highest communicative abilities and participant treatment fidelity. By contrast, Niko and Peter required additional reinforcement contingencies to increase sleep-conducive behavior. Individuals with more severe intellectual disability may struggle to refrain from engaging in sleep-interfering behaviors that offer immediate reinforcement and may experience difficulty generating appropriate alternative responses (Ho et al. 2015). Niko attempted to gain access to electronic devices as intervention began, only engaging in sleep facilitative behavior after access had been restricted. Although Eric evidenced relaxation training mastery, Peter required significant support to learn relaxation skills (e.g., social story, visual aids, modeling). After four sessions, his relaxation technique still did not appear correct and he was not able to identify when to utilize the strategies. Echolalia and compromised memory and sequencing abilities appeared to limit Peter's ability to engage in conversation. Niko and Peter also each had difficulty attending to therapeutic tasks. Adolescents with ASD are less likely to initiate social interaction (Chevallier et al. 2012). Limited social skills may have contributed to the inhibited engagement with Peter and Niko.

Parents within the current study consistently reported that adolescents had become responsible for their own sleep and had developed skills to engage in appropriate sleep behavior independently, likely facilitating the maintenance of treatment effects over an extended time period. Psychoeducation may

have been sufficient for Eric to reduce use of electronic devices without resistance, particularly as this was not the primary maintaining reinforcer for his sleep-interfering behavior. Furthermore, an increase in Peter's sleep-conducive behavior (e.g., eyes closed, lying still) was observed following explicit, concrete instructions within a social story. Interestingly, he also began reprimanding other family members for sleeping during the day. Parents play an important role enforcing limits to facilitate sleep-conducive behavior and delivering treatment directly to adolescents provides them with the skills and knowledge to sustain healthy sleep practices. Treatment maintenance is critical to help prepare adolescents with ASD for adulthood and ensure they can function most effectively within living, education, and vocational settings without being compromised by the effects of sleep disturbance.

Parent treatment fidelity was an issue for one of the three families. Eric's parent completed every component of the treatment plan on 26% of nights. It was particularly difficult for this family to refrain from providing social attention post-bedtime and to maintain consistent sleep and wake times respectively. In this case, low parent treatment fidelity did not appear to affect Eric's sleep outcomes, perhaps because his own treatment fidelity was high. Treatment fidelity was not a challenge for Niko and Peter's parents. The complexity of everyday life for parents of children with neurodevelopmental disorders may impact their ability to consistently focus their attention and energy on improving their child's sleep and prioritize this one aspect of their lives (Beresford et al. 2016).

Videosomnography has rarely been used within the ASD and sleep literature; however, this objective measure can be collected within participants' homes and enables detection of salient information (e.g., topographies of sleep and awake behavior) unobtainable through actigraph or polysomnography recordings (Moore et al. 2017). High IOA between parent-reported sleep diaries and videosomnography was found, replicating results of previous sleep research with young people with ASD (Jin et al. 2013). However, our videosomnography data revealed parents were often unable to detect covert sleep-interfering behavior (e.g., early morning device use). As a result, they struggled to identify the length of sleep onset or wakings. Subjective sleep measures may be more reliable within pre-school and school-aged populations, as sleep-interfering behavior tends to involve overt signaling to the parent or significant disruption to the household. Understandably, as Eric's case suggests, increasing desire for privacy may prevent adolescent participants consenting to the use of videosomnography. Further, increasing sexual desire during adolescence, paired with the lack of social understanding apparent in many individuals with ASD, may put this population at risk of being inadvertently recorded while engaging in sexual activities. Privacy issues necessitate careful consideration of videosomnography with any population, but particularly perhaps with adolescents.

Limitations and Future Directions

The current study illustrates treatment components directed towards adolescents are viable and may be beneficial additions to traditional parent-mediated sleep interventions when working with verbal adolescents with ASD. Future research aimed at determining the extent to which intellectual functioning and motivation may influence the effectiveness of sleep interventions implemented with adolescents appears warranted. Of consideration within the current study is the small number of participants with heterogeneous presentations of sleep problems and ASD characteristics, inhibiting generalizability to other adolescents with ASD. Second, recording covert sleep-interfering behavior via parent report may not be sufficiently accurate, suggesting a need for research aimed at evaluating the veracity of dependent variables used in sleep-intervention research. Third, the single-case design used did not isolate the effects of any specific intervention component and it is not possible to determine whether any given component was necessary or sufficient. However, the results of this pilot study suggest future research could include a component analysis and experimental design capable of demonstrating experimental control.

Author Contributions JvD: designed and executed the study, conducted data analyses, and wrote the paper. LM and KM contributed equally towards the collaboration of the design, execution, and writing of the study. NB: collaborated with the data analysis and writing of the study. RL: collaborated with the writing of the study. JH: assisted with clinical application of assessment and intervention to Peter.

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Compliance with Ethical Standards

Conflict of Interest The authors declare that they have no conflict of interest.

Ethical Approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the University of Canterbury Human Ethics Committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed Consent Informed consent was obtained from all participants.

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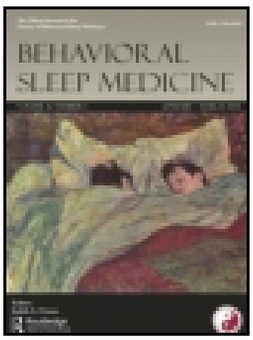
Appendix W: van Deurs, J. R., McLay, L. K., France, K. G., & Blampied, N. M. (2020). Sequential implementation of functional behavioural assessment-informed treatment components for sleep disturbance in autism: A case study. Behavioral Sleep Medicine. Advance online publication. doi:10.1080/15402002.2020.1758701



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Sequential Implementation of Functional Behavior Assessment-Informed Treatment Components for Sleep Disturbance in Autism: A Case Study

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ABSTRACT

Background: Sleep disturbances are a significant problem for people with autism spectrum disorder (ASD). Existing research supports the use of parent-implemented, functional behavior assessment (FBA)-informed interventions for sleep problems in children with ASD. There is also emerging evidence for combined parent- and young person-implemented behavioral sleep interventions for older children and adolescents with ASD. However, the active treatment components of such interventions have not been identified in previous studies, as components have not been evaluated independently of one another.

Methods: The current study sequentially implemented FBA-informed treatment components (in the order of least to most restrictive and time intensive) within a single-case AB design, to evaluate at which point treatment resulted in a statistically and clinically substantive reduction in target sleep variables. Combined parent- and young person-implemented intervention components consisted of: (a) white noise; (b) white noise and relaxation instruction; and (c) white noise, relaxation instruction, and stimulus control.

Participant: The participant was a 9-year-old girl with autism and selective mutism.

Results: The combined use of white noise, relaxation instruction, and stimulus control resolved the participant's sleep problems. Other more restrictive and/or time intensive interventions were unnecessary. Treatment effects were maintained at 10-week follow-up.

Conclusions: The current study illustrates the feasibility of administering FBA-informed treatment components sequentially, to ensure application of minimally sufficient interventions.

The Autism and Developmental Disabilities Monitoring (ADDM) Network estimates 1 in 59 children in the United States have autism spectrum disorder (ASD; Baio et al., 2018); a neurodevelopmental disorder characterized by difficulties with social communication and restricted/repetitive thoughts and behavior patterns. In addition to these two core features, the neurological functioning of people on the autism spectrum can compromise their speech and language skills, sensory responsivity, executive functioning (e.g., emotional and behavioral regulation, attention), and psychological wellbeing (e.g., anxiety). Sleep disturbance is also a significant

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This article is an expansion of previous work and contains re-use of some material within the methods section of van Deurs, J. R., McLay, L. K., France, K. G., Blampied, N. M., Lang, R. B., & Hunter, J. E. (2019). Behavioral sleep intervention for adolescents with autism spectrum disorder: A pilot study. *Advances in Neurodevelopmental Disorders, 3*, 397–410. doi:10.1007/s41252-019-00123-z.

Supplemental data for this article can be accessed on the publisher's website.

clinical problem faced by many people on the autism spectrum. Rates of sleep disturbance in children and adolescents with ASD range from 50% to 80% in studies conducted in the USA and Canada (Couturier et al., 2005; Krakowiak et al., 2008; Malow et al., 2016; Souders et al., 2009). Difficulty initiating and maintaining sleep, as evidenced by prolonged sleep latency, shorter sleep duration, night wakings, and reduced sleep efficiency (time spent asleep in bed compared with total time in bed), are the most common sleep problems experienced by young people with ASD (Herrmann, 2016). These issues tend to persist throughout childhood and adolescence if untreated (Sivertsen et al., 2012), although the phenomenology changes with age (Goldman et al., 2012). Parents of children with ASD report higher rates of bedtime resistance, night wakings, parasomnias, and sleep anxiety, whereas parents of adolescents with ASD report higher rates of sleep onset delay, shorter sleep duration, and daytime sleepiness (Goldman et al., 2012).

Sleep deprivation exacerbates the difficulties already faced by young people on the autism spectrum, further compromising their cognitive and adaptive functioning, behavior, and emotional wellbeing (Nadeau et al., 2015; Park et al., 2012; Sikora et al., 2012). For example, mild ASD severity is typically associated with lower levels of behavior problems than higher ASD severity, however, young people with ASD and sleep disturbance are likely to exhibit clinical levels of problem behavior, regardless of ASD severity (Lindor et al., 2019). Further, the effects of sleep disturbance are not isolated to the individual but can also affect family functioning. Parents of young people with ASD and sleep problems report higher stress and poorer mental health than parents of young people with ASD without sleep problems (Martin et al., 2019).

Bidirectional interactions between physiological (e.g., dysregulated melatonin), behavioral (e.g., operant conditioning of sleep-interfering behaviors), and psychological (e.g., comorbid psychopathologies) factors contribute to the etiology of sleep problems in people with ASD. While pharmacological sleep interventions (e.g., prescribed melatonin) can address underlying physiological contributions, they neglect other factors precipitating and maintaining the sleep problem. Growing evidence supports the use of parent-implemented behavioral interventions for the treatment of sleep disturbance in children with ASD (Carnett et al., 2019). There is also emerging evidence for the use of combined young-person- and parent-implemented cognitive behavioral interventions for older children and adolescents with ASD (Loring et al., 2016; McCrae et al., 2019; Van Deurs et al., 2019).

Functional behavior assessment (FBA; Newcomer & Lewis, 2004) is an assessment procedure commonly used to identify antecedent and consequence variables that may have established and may now be maintaining target behavior. FBA involves analyzing recurring patterns among antecedent events, behaviors, and consequences, based on information obtained from a range of sources (e.g., interviews, questionnaires, behavior diaries). Functional assessment procedures have been shown to maximize the efficiency and effectiveness of behavioral interventions and are commonly used to inform treatment of general behavior problems in young people with ASD (Heyvaert et al., 2014). A number of studies highlight the importance of using FBA to inform sleep interventions for children with ASD (e.g., Jin et al., 2013; McLay et al., 2019, 2018) and indicate that similar behavioral topographies (e.g., leaving the bedroom post-bedtime) do not necessarily serve the same function, nor warrant similar treatment. For one child, sleep-interfering behavior may be maintained by parent attention, and for another may involve salient but inappropriate controlling stimuli (e.g., electronic devices). Disambiguating such alternative functional relationships is critical to the development of targeted, effective interventions.

A recent review of behavioral interventions for sleep disturbance in children with ASD revealed all existing studies have consisted of multiple components (Carnett et al., 2019). Consequently, the specific treatments responsible for behavior change are unknown, and as the effects of each component have not been evaluated independently from one another, it is unclear which strategies are necessary and minimally sufficient to produce change for any one individual. Furthermore, treatment is less likely to be adhered to when it is disruptive to family routine, time intensive, complex, or evokes distress. This is concerning given comprehensive FBA-informed treatment packages can be challenging to deliver and

involve extensive time commitment. The minimal sufficiency principle emphasizes the implementation of treatment components which balance effectiveness with ease of delivery (Sanders et al., 2014). Thus, the identification of minimally sufficient methods (i.e., those which are less restrictive and time-consuming) to address the function of sleep-interfering behavior may be critical to facilitating treatment fidelity and maintenance of behavior change. Although there are no specific guidelines outlining the least to most restrictive applied behavior analytic interventions, antecedent-based interventions and reinforcement are generally perceived to be less aversive or drastic than punishment techniques (e.g., extinction; Bailey & Burch, 2013; Kazdin, 2013).

A range of objective and subjective measures can be used to assess sleep-related problems, with each capturing unique phenomena within the same construct. Sleep diaries gather continuous data on sleep patterns for a period of time, whereas questionnaires require informants to make retrospective judgments about sleep patterns over extended time periods and may be at increased risk of recall bias. Research indicates there may be discrepancies between information obtained from different measures. For example, parents are simply not able to detect covert wakings where their child remains quiet in bed, evident via video observation. Parents and adolescents are reliable informants of overt variables, such as bedtime and waketime, but they have difficulty evaluating covert variables, such as duration of sleep onset or night wakings (Bauer & Blunden, 2008). Videosomnography has been used rarely within the ASD and sleep literature. Additionally, self-report sleep diaries (commonly completed by typically developing young people) have been scarcely used by young people with ASD. Further, no single study within the ASD and sleep literature has compared information obtained from parent-report sleep diaries, self-report sleep diaries, videosomnography, and questionnaire outcomes.

The aims of the present study were to (a) sequentially administer, minimally intrusive, FBA-informed treatment components in an attempt to resolve sleep disturbance in a child with ASD; (b) examine parent- and child-reported social validity of treatment approaches; and (c) compare information gathered via parent-report sleep diaries, self-report sleep diaries, videosomnography, and questionnaires.

Method

Participant

Eve (pseudonym) was a 9-year-old girl with autism and selective mutism (evidenced by her failure to speak in specific situations not attributable to her speech and language abilities). The Communication sub-domains of the Vineland Adaptive Behavior Scales Third Edition, Comprehensive Parent/Caregiver Form (Vineland-3; Sparrow et al., 2016) revealed Eve had below average receptive and expressive language abilities, equivalent to a 2 year, 9-month-old child, and a 3 year, 11-month-old child, respectively. She had high reading and writing skills, equivalent to an 8 year, 6-month-old child. Analysis of Vineland-3 items and clinical assessment (interviews and observation) revealed Eve could ask and answer questions that involved *when* and *why*, she could sometimes describe everyday events in detail, write, or draw instructions for others, fill out forms with more than two pages, and attend to and understand a 30-min informational talk. Eve was referred to the study by her parents who reported that she took an extended period of time to initiate sleep at bedtime (60– 120 min), engaged in multiple curtain calls (CCs, bids for parental attention post-bedtime) and woke every night, occasionally (up to three times a week) for several hours. Eve received 2.5 mg of slow-release melatonin nightly, which was prescribed by her physician.

Design

This study employed an AB single-subject design whereby the intervention phase consisted of sub-phases with cumulative addition of treatment elements until a sufficient treatment response was

observed. Treatment effects were replicated across five target sleep variables, including CCs, SOL, NWs, total sleep duration, and sleep efficiency.

Setting

Pre- and post-treatment assessment interviews and treatment planning discussions were conducted in the family home. This reduced travel burden for the family, likely preventing dropout, and contributed to a greater understanding of Eve's sleep environment. Treatment was implemented within the home by Eve and her parents with support from a psychologist (JvD). During treatment, communication with Eve's parents was conducted in person, or via telephone, and e-mail. As Eve experienced high anxiety speaking on the telephone, the psychologist communicated with her face to face and via letters using language appropriate for her reading level.

Measures

Clinical interviews

A semi-structured interview was conducted separately, with Eve and her mother. Information was gathered regarding Eve's interests and strengths; developmental history; current and historic sleep concerns; factors contributing to sleep disturbance (e.g., Eve's thoughts and emotions at bedtime); motivation to improve sleep; and family context. To facilitate communication with Eve the interview included visual stimuli (e.g., sleep cartoons); drawing and written activities; incorporation of Eve's special interests; metaphors (e.g., magic wand to address sleep disturbance); and open, closed, and multiple-choice question format. Eve was given extended time to answer questions and typically did so within 10 to 20 s.

Sleep Assessment Treatment Tool (SATT; Hanley, 2005)

The SATT was incorporated in the clinical interview with Eve's mother. The SATT is a semi-structured interview used to identify factors underlying children's sleep disturbance.

Questions About Behavior Function (QABF; Matson & Vollmer, 1995)

The QABF was administered to Eve's mother following the clinical interview (enabling further explanation of items as needed). The QABF is a 25-item rating scale used to establish the function (e.g., social attention, escape, tangible reinforcement) of a target behavior.

Multidimensional Anxiety Scale for Children 2nd Edition (MASC 2; March, 2012)

The MASC 2 was completed by Eve and her parents during assessment to measure existing anxiety levels. The MASC 2 is a 50-item multi-informant questionnaire designed to assess anxiety symptoms (e.g., Social Anxiety, Separation Anxiety, Physical Symptoms) experienced by people aged 8 to 19 years. The number of elevated scores across Anxiety Scales yield an Anxiety Probability score, classified as Low, Borderline, High, or Very High.

Children's Sleep Habits Questionnaire (CSHQ; Owens, Spirito, McGuinn, 2000)

The CSHQ was completed by Eve's parents during the assessment and maintenance phase to evaluate the change in parent-reported sleep disturbance. The CSHQ is a widely used parent-report questionnaire for assessing school-aged children's sleep patterns according to the frequency of specific behaviors across eight sleep domains (Bedtime Resistance; Sleep Onset Delay; Sleep Duration; Sleep Anxiety; Night Wakings; Parasomnias; Sleep Disordered Breathing; and Daytime Sleepiness) within a typical week. Higher scores are indicative of poorer sleep, and scores ≥41 are indicative of clinically significant sleep disturbance (Owens, Spirito, McGuinn, 2000). The CSHQ has satisfactory internal consistency (0.68 to 0.78) and test–retest reliability (0.62 to 0.79; Owens, Spirito, McGuinn, 2000).

The Sleep Self-Report (SSR; Owens, Spirito, McGuinn, & Nobile, 2000)

The SSR was completed by Eve during the assessment and upon entering the maintenance phase as a measure of child-reported sleep pre- and post-treatment. The SSR is a 26-item self-report questionnaire for children 7 to 12 years of age which corresponds with the CSHQ subscales (Owens, Spirito, McGuinn, & Nobile, 2000). Eve rated the frequency of specific sleep behaviors engaged in over the past week on a 3-point Likert scale (Owens, Spirito, McGuinn, & Nobile, 2000). SSR scores have a possible range of 23 to 69 with higher scores indicative of worse sleep. The SSR has adequate discriminative validity and test–retest reliability (0.76– 0.88; Orgilés et al., 2013; Owens, Spirito, McGuinn, & Nobile, 2000; Steur et al., 2019).

Sleep diaries

Eve's parents completed daily sleep diaries during each phase of the study. Recorded information included the (a) frequency and duration of daytime sleep; (b) duration of sleep onset latency (SOL); (c) frequency of CCs; (d) frequency and duration of night wakings (NWs); and (e) time of morning waking. Participant's sleep setting, behavior during CCs and NWs, and parents' responses to this behavior were also noted. Eve was also asked to complete daily sleep diaries which included the addition of the type and intensity of emotions experienced before bedtime.

Videosomnography

A D-Link HD Cloud Camera was used to directly observe and measure Eve's sleep during each study phase. The camera was placed at the end of Eve's bed and was set to turn on at her typical bedtime and switch off at her typical rise time. Information obtained from the video included the same information reported in sleep diaries, plus the addition of topographies of awake behavior post-bedtime (e.g., vocalizations, play) and topographies of sleep behavior (e.g., sleep position, eye movement, limb movement). The following operational definitions were used to code video (a) asleep, lying down with minimal non-discrete movement for ≥5 min, and no indication of wakefulness; and (b) awake, the presence of any sleep-interfering behavior, eyes open, or frequent physical movement (Jin et al., 2013). Video footage was coded by a researcher blind to parent sleep diary recordings and enabled objective monitoring of Eve's progress and the collection of interobserver agreement data.

Interobserver agreement (IOA)

Agreement between parent- and self-report diary data and video observations was calculated. Sleep phenomena which parents could not be expected to detect (e.g., covert awakenings in which Eve remained quiet in her bed) were omitted from IOA calculations. IOA for CC frequency was calculated on occasions whereby bids for attention were detectable by video observers (e.g., clear calling out). Measures of duration (e.g., SOL) and sleep/wake times were considered in agreement if they were ± 15 min. Percent agreement for target behaviors was calculated using the equation [Agreement/(Agreement + Disagreement)] × 100. IOA data were collected for 35% of nights across all study phases.

Treatment fidelity

Treatment fidelity was assessed on 88% of nights across intervention and follow-up phases by comparing measurable events recorded in contact notes, video footage, and sleep diaries with the prescribed treatment protocol. An aggregate treatment fidelity score was calculated using the formula (Completed tasks/Total tasks) × 100.

Social validity

Eve completed the Young Person Treatment Evaluation (YPTE) post-treatment to assess her perception of the intervention. The YPTE, developed by the authors based on the Child Evaluation Inventory (CEI; Kazdin, 1984), consists of six items assessing effectiveness, enjoyability,

fairness, time required, and overall perception, using a 3-point Likert scale (e.g., 1 = Not at all helpful; 2 = OK; and 3 = Very helpful). Ratings are summed to provide a total acceptability score. Post-treatment, Eve's parents completed the Treatment Acceptability Rating Form-Revised (TARF-R; Reimers et al., 1992), a 20-item questionnaire based on the adult version of the CEI. Ratings on six subscales (Effectiveness; Reasonableness; Willingness; Cost; Negative side-effects; Disruption/time) are summed to provide a total treatment acceptability score. Post-treatment interviews were also conducted with Eve and her parents individually to further evaluate social validity and gather qualitative information regarding treatment effects.

Procedure

FBA

Information obtained from the SATT, the QABF, sleep diaries, and analysis of video footage were used to conduct the FBA and synthesized in an FBA-informed case conceptualization which then guided treatment planning (Blampied, 2013). FBA indicated that many antecedent and consequence variables were contributing to Eve's sleep disturbance. Table 1 describes the factors hypothesized to precipitate and maintain each of Eve's sleep difficulties as well as their function.

Assessment revealed Eve's bedtime varied between 8:00 and 9:00pm. Once in bed, Eve played games on an iPad, listened to podcasts, or read using an e-reader until she fell asleep. Her parents tended to bid her goodnight and switch off the light at variable times during this period, following which Eve continued to engage in sleep-interfering activities. Eve's bedroom was situated close to the main living areas and she kept her bedroom door open at night; she complained the television volume was too loud post-bedtime. Parent ratings on the MASC 2 yielded Total Anxiety scores within the Very Elevated range and indicated a Very High Probability of an anxiety disorder. This was corroborated by self-report ratings on the MASC 2. Additionally, Eve reported experiencing frequent and intense worry about her parent's wellbeing and consequently regularly listened to and monitored her parents' conversations post-bedtime, interjecting at times. Further, she typically called out to her parents multiple times to request food or drink, and discuss other worries. Her parents responded inconsistently, providing comfort/reassurance and/or requested items, or reprimanding the behavior. Eve usually woke multiple times per night for no identifiable reason or in response to her baby sibling crying, then experienced difficulty reinitiating sleep. These wakings generally resulted in her using her e-reader. On some occasions Eve did not return to sleep at all.

Antecedent variables hypothesized to be implicated in Eve's sleep disturbance included lack of physiological sleep pressure, lack of consistent discriminative stimuli (events which signal the availability of the reinforcer [sleep onset], such as the bedroom light being switched off) for sleep, salient discriminative stimuli for sleep-competing behavior (e-reader), external noise, and cognitive and physiological arousal. Sleep-interfering behavior was thought to be reinforced by social attention, access to tangibles (e-reader), and escape from distressing cognitions.

Table 1. Factors precipitating and maintaining sleep disturbance, hypothesized function, and treatment components.

	Curtain calls	Delayed sleep onset	Frequent and extended night wakings
Factors thought to be precipitating and/or maintaining behavior	Lack of physiological sleep pressure; parent responses to curtain calls; cognitive and physiological hyperarousal	Lack of physiological sleep pressure; lack of discriminative stimuli for sleep; salient discriminative stimuli for sleep-competing behavior (e-reader); loud external noises; cognitive and physiological hyperarousal	Lack of physiological sleep pressure; lack of discriminative stimuli for sleep; salient discriminative stimuli for sleep-competing behavior (e-reader); loud external noises; cognitive and physiological hyperarousal
Hypothesized function	Social attention Access to tangibles Escape (from distressing cognitions)	Social attention Access to tangibles	Access to tangibles

Baseline

Baseline commenced following completion of the FBA. Eve was randomly assigned a baseline length of 4 weeks (from a possible pre-specified range of 2 to 4 weeks to reduce the possibility of a biased selection by the researcher). Her family were asked to maintain existing sleep habits during this phase.

Intervention

Intervention commenced immediately following baseline. FBA-informed intervention components were implemented sequentially across phases until Eve's sleep disturbance resolved. Treatment components were selected according to those which were hypothesized to most appropriately address the function of the behavior and ordered from the least to most restrictive and time intensive. Up to five intervention phases were planned; treatment proceeded to the next phase if clinically substantive improvement across target sleep variables had not occurred within 7 days. Eve entered maintenance phase (continuation of the final treatment protocol without therapist input or data collection) when there was a clinically substantive reduction in target behaviors lasting 14 days.

Given Eve's difficulty engaging with new people, her parents were responsible for providing psychoeducation and introducing her to therapeutic techniques, following coaching from the psychologist. They were contacted daily to weekly by the psychologist to facilitate effective treatment delivery and monitor fidelity. Eve had therapist contact at one to two weekly intervals via letters and certificates of achievement.

Treatment phase 1: White noise (night 30 to 40). The first phase of treatment involved playing two preferred sounds, selected by Eve (cat purring and fire crackling) from the Rain Rain application (https://www.rainrainapp.com/) from bedtime to morning wake time, and her door was also closed to mask external noises (e.g., baby sibling crying, television). These strategies inhibited excessive reassurance seeking as Eve could no longer hear, monitor, and respond to adult conversation from her bed (Eve's parents were not given any instructions regarding how to respond to CCs should they occur). Further, the white noise may have acted as a salient proximate discriminative stimulus for sleep. Eve and her parents chose a volume which masked external noise while remaining at a comfortable level.

Phase 2: White noise and relaxation instruction (night 41 to 51). On night 41, Eve was taught relaxation strategies (diaphragmatic breathing and progressive muscle relaxation) to provide her with skills to independently alleviate hyperarousal and facilitate independent initiation of sleep at bedtime and during wakings. She was taught the relaxation strategies during one in-home session with a psychologist who used modeling and incorporated Eve's soft toys in the instruction, aided by a short story which included pictures of Eve's favorite animal (cat) completing the exercises. As well as continuing to use white noise, Eve's parents were instructed to read the book with Eve each night at bedtime and guide her through the relaxation exercises.

Phase 3: White noise, relaxation instruction, and stimulus control (night 52 to 72). During the third intervention phase, Eve's sleep was brought further under appropriate stimulus control; both interoceptive (e.g., tiredness) and exteroceptive (e.g., consistent bedtime) discriminative stimuli for sleep were strengthened, and dependencies non-conducive to sleep were eliminated. Bedtime fading and a consistent sleep/wake schedule ensured Eve had sufficient homeostatic sleep pressure to initiate sleep quickly once in bed. She was instructed to use her bed for sleep only and e-reader use restricted to when she was seated at her desk up until bedtime. Eve's parents instructed her to stop using the e-reader at a consistent time each night and bid her goodnight once she was in bed, providing clear sleep cues (once again Eve's parents were given no instruction regarding how to respond to any CCs). These arrangements functioned to make the bed a discriminative stimulus for sleep. To reinforce Eve for no longer using her e-reader in bed, she was allowed to stay up with her parents in the lounge and use her e-reader until bedtime during the first week of Phase 3.

Phase 4: White noise, relaxation instruction, stimulus control, and reinforcement. The planned fourth intervention phase consisted of parent-delivered positive reinforcement (e.g., a small tangible reward) contingent on Eve's engagement in sleep-conducive behavior (e.g., remaining in bed post-bedtime). Eve's parents would not have received instruction regarding their responses to any CCs.

Phase 5: White noise, relaxation instruction, stimulus control, reinforcement, and unmodified extinction. In order to address the attentional component hypothesized to underlie Eve's sleep disturbance, the planned fifth intervention phase involved eliminating inadvertent social reinforcement for sleep-interfering behavior. Eve's parents would have been instructed not to attend to any bedtime disruptions (e.g., calling out). If Eve left her bedroom, her parents would have been required to return her to bed with minimal engagement.

Follow-up

Follow-up video and sleep diary data were collected for 7 nights at 10 weeks post-treatment.

Data analyses

Visual analysis (examination of level, trend, and stability) of graphed data supplemented by percentage of data points below/exceeding the baseline median (PBM/PEM; Ma, 2006), was used to evaluate the effect of each treatment phase on target behavior. PBM/PEM is an effect-size measure for single-case data and can be interpreted as follows: <70% represents ineffective treatment; 70% to 90% moderate effectiveness; and >90% high effectiveness (Ma, 2009). Within the current study, the baseline median for each target behavior was compared to the individual data points within each treatment phase.

To indicate whether improvement in target variables was clinically substantive, Eve's baseline and treatment data were compared to developmental norms. Clinical cutoffs are presented in Figures 1–5 to discriminate between appropriate and poor sleep quality. Indicators of good sleep quality in typically developing school-aged children include: <30 min SOL, ≤1 waking, ≤20 min of wake-after-sleep-onset (WASO; duration of NWs), and ≥85% sleep efficiency (Ohayon et al., 2017). Conversely, indicators of poor sleep quality or clinical sleep problems for this age group include: >45 min SOL, ≥4 wakings (at least 5 min in length), >40 min of WASO, and <75% sleep efficiency (Ohayon et al., 2017). Optimal sleep duration for school-aged children is between 9 and 11 hours; 7 to 8 hours are considered appropriate for some children, and less than 7 h (420 min) are not recommended (Hirshkowitz et al., 2015). The current study applied a 7-h total sleep time clinical cutoff. As appropriate levels of CCs have not been described within the literature, the current study defined ≥1 CCs more than 5 times a week as a clinical problem.

Pearson product-moment correlations (r) were calculated to measure the strength of the relationship between parent-report sleep diaries, self-report sleep diaries, and video observations. Following Cohen (1988), $r > 0.50$ represents a strong relationship, 0.30 to 0.49 a medium relationship, and <0.30 a small relationship between variables.

Results

Agreement between sleep measures

Mean IOA between parent-report diaries and video was 89% (range, 57–100%) across study phases and between self-report diaries and video was 36% (range, 0–100%) across study phases. IOA values across target variables and study phases are presented in Supplementary Material Tables 1 and 2.

Strong positive correlations were found between data obtained from parent-report sleep diaries and video data for SOL, CCs, and sleep efficiency (range, $r = 0.80$–0.82; see Supplementary Material Table 3 to 7). Smaller correlations were found between parent-report and video data for duration of NWs and total sleep duration (range, $r = 0.56$–0.57), although they exceeded Cohen's convention for

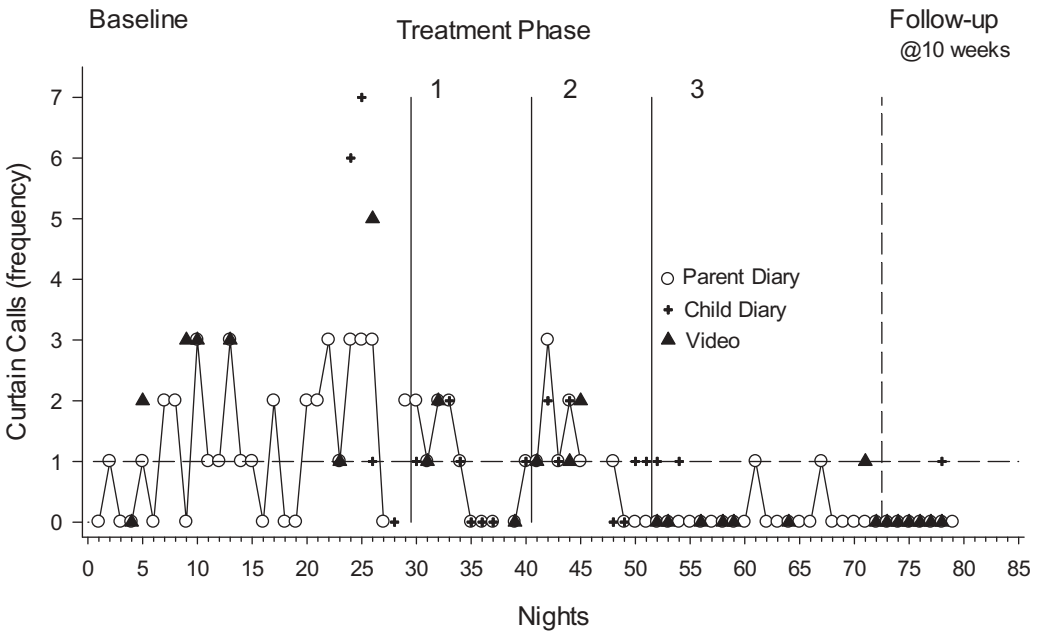

Figure 1. Frequency of Eve's curtain calls across baseline, intervention, and follow-up phase.

Note. The dashed horizontal line is the cutoff for clinical levels of curtain calls (≥1) engaged in by school-aged children.

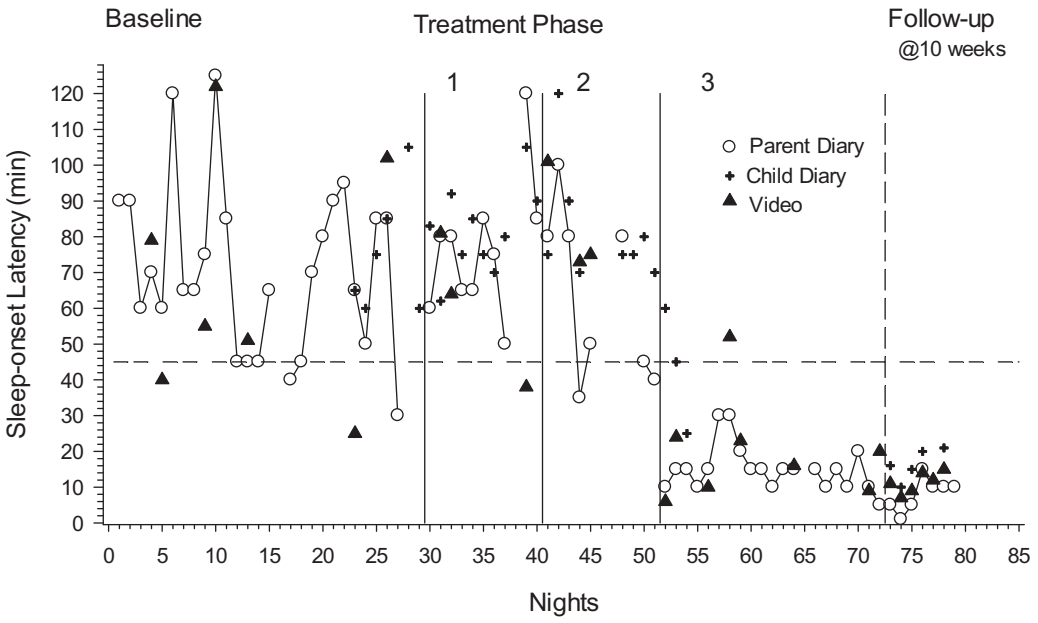

Figure 2. Eve's sleep onset latency across baseline, intervention, and follow-up phase.

Note. The dashed horizontal line is the clinical cutoff for sleep onset delay (>45 min) indicative of poor sleep in school-aged children.

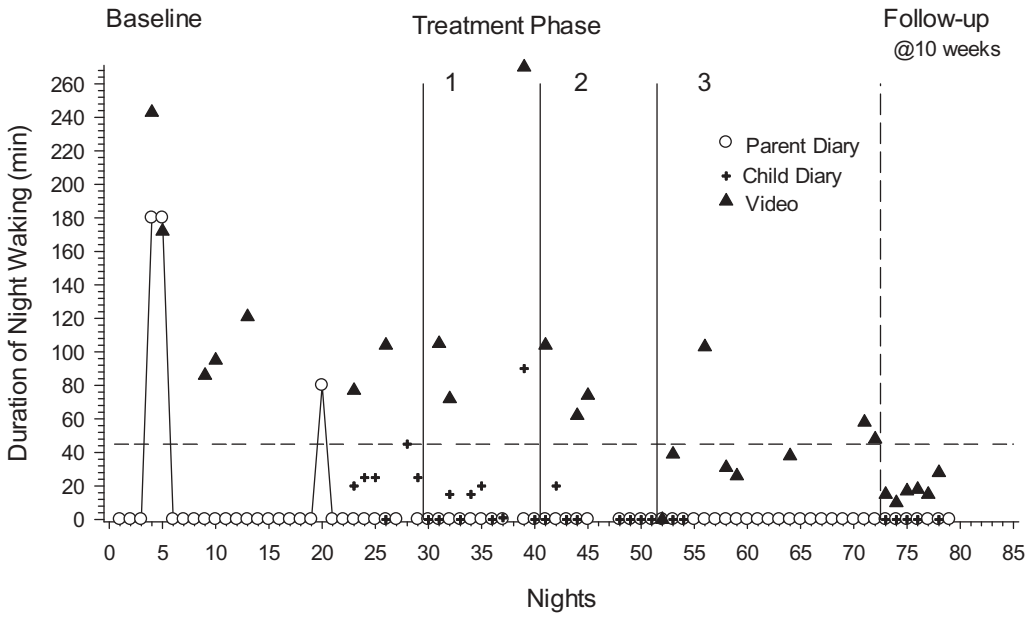

Figure 3. Duration of Eve's night wakings across baseline, intervention, and follow-up phase.

Note. The dashed horizontal line is the clinical cutoff for duration of night wakings (>40 min) indicative of poor sleep quality in school-aged children.

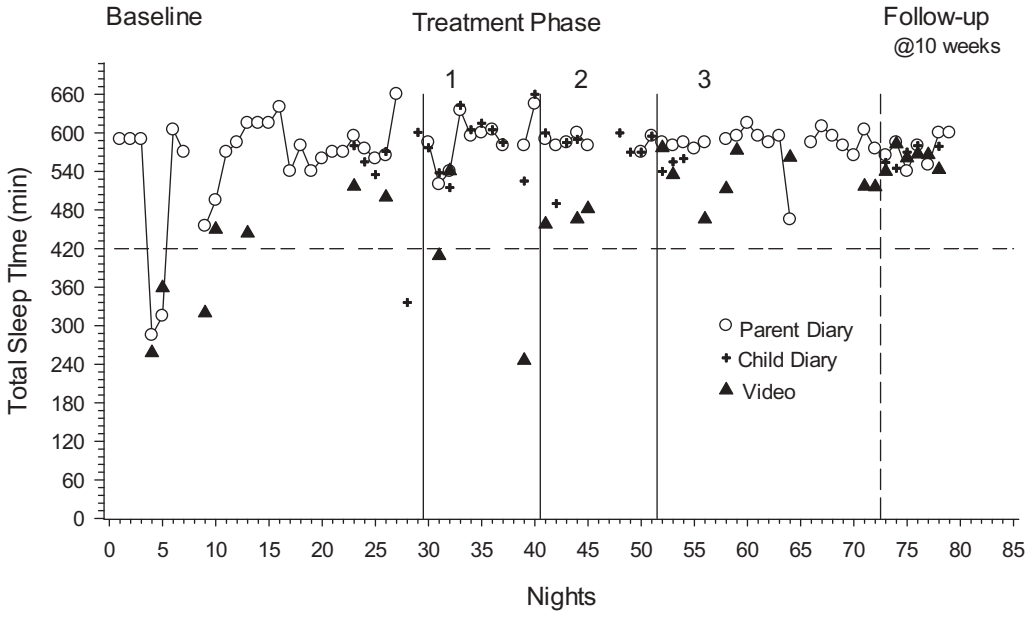

Figure 4. Eve's total sleep time across baseline, intervention, and follow-up phase.

Note. The dashed horizontal line is the clinical cutoff for poor sleep duration (420 min) in school-aged children.

a large relationship. Strong positive correlations were found between self-report sleep diaries and video data for SOL, duration of NWs, and sleep efficiency (range, r = 0.68–0.85). Small to moderate correlations were found between self-report and video data for CCs and total sleep duration (range,

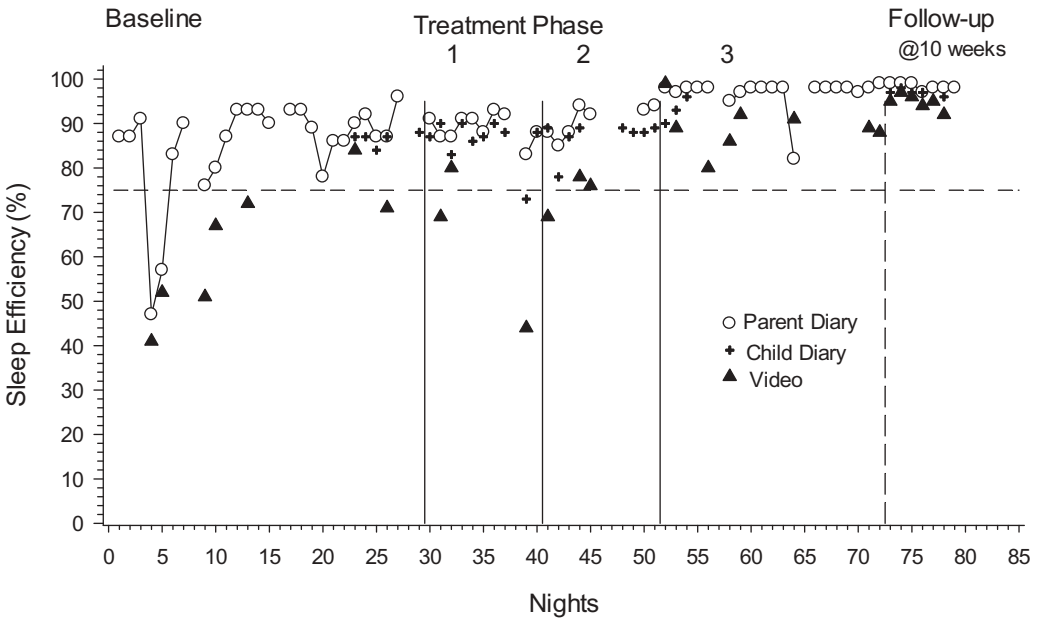

Figure 5. Eve's sleep efficiency across baseline, intervention, and follow-up phase.

Note. The dashed horizontal line is the clinical cutoff for poor sleep efficiency (<75%) in school-aged children.

r = 0.21–0.48). Parent-report and self-report diary data were strongly correlated across all target sleep variables (range, r = 0.72–0.87).

The correlations indicate that parent-report, self-report, and video data followed a similar pattern over time (e.g., reduction in sleep problems). However, there were large differences between individual numerical values reported at specific time points across video and parent- or self-report sleep diaries.

Data quality

As Eve's parents could not always detect covert wake behavior (hence exclusion from IOA analysis) there were discrepancies between parent-report and video observed values across target behaviors. For this reason, video observations are reported to supplement parent-report. Eve completed self-report sleep diaries from night 23 to 54. These discrepancies notwithstanding, parent-report is the primary dependent variable reported, since there was more continuous data from this source than from the other sources.

Necessary treatment components

Eve met criterion for maintenance during the third phase of treatment (consisting of white noise, relaxation instruction, and stimulus control), therefore additional planned intervention phases involving reinforcement and extinction procedures were not implemented. Overall, intervention lasted for 43 days.

Effect on CCs

Parent-reported CCs ranged from none to three per night in baseline, with most baseline nights having one CC. There was notable variance in CC levels across parent-report, self-report, and video

(parent-report range, 0– 3; self-report range, 0– 7; video range, 0– 5). There was some reduction in CCs in the first treatment phase, but this was not maintained in the second phase. Only with the full combined treatment (Phase 3) was there consistent reduction in CCs to zero (or one per night at worst), and this was maintained at follow-up. The reduction in CCs achieved was clinically substantive, and stable (Phase 3 parent-report PBM = 91%, video PBM = 100%; follow-up parent-report PBM = 100%, video PBM = 100%).

Effect on SOL

Eve consistently experienced clinical levels (>45 min) of SOL during baseline (parent-report median = 67.5 min, self-report median = 70 min, video median = 55 min), with a substantial number of nights having clinically severe levels (>90 min). There was little evidence of change in SOL during treatment Phase 1 or 2; only in Phase 3 was a clinically substantive reduction in SOL observed (parent-report, self-report, and video PBM = 100%). The reduction in median SOL from baseline to the final phase of treatment was −53 min (parent-report), −25 min (self-report), and −37 min (video). SOL reduction was maintained at follow-up (parent-report, self-report, and video PBM = 100%) as Eve fell asleep within 15 min each night.

Effect on NWs

Parent-report and self-report indicated the duration of Eve's NWs was not of clinical concern (i.e., >40 min) during baseline or at any point thereafter, but video recordings showed Eve woke regularly during baseline for extended time periods (range, 77– 243 min). Video data show that there was an immediate, clinically significant reduction in the duration of NWs only during Phase 3 (video PBM = 100%). The median video duration reduced from 104 min in baseline to 39 min in the final phase, however, the length of WASO was still of clinical concern on occasional nights during this phase. Video data demonstrated there was a further reduction in the duration of NWs during follow-up (video median = 16 min) and length was not of clinical concern on any night.

Effect on total sleep time and sleep efficiency

Video data showed Eve's total sleep duration (video median = 444 min) was more problematic than parent-report or self-report indicated (parent-report median = 572.5 min, self-report median = 563 min), with a number of nights in baseline within the clinical severity range (<420 min). Nevertheless, there was still an improvement in parent-reported sleep duration within each treatment phase (parent-report PEM Phase 1 = 90%, Phase 2 = 86%, Phase 3 = 91%). Video data revealed both Phases 2 and 3 had large treatment effects (video PEM Phase 2 and 3 = 100%), which were clinically substantive, with median sleep duration increasing from 444 min in baseline to 525 min in the final phase. In addition, video data revealed Eve reached optimal sleep duration (540 to 660 min/9 to 11 hours) on occasion during treatment. Improvement in total sleep duration was maintained at follow-up (video median = 564 min) and Eve achieved optimal duration of sleep each night.

Video data showed Eve had poor sleep efficiency during baseline (video median = 67%), although parent- and self-report suggested it was less problematic (parent-report and self-report median = 87%). Both parent-report and video data indicated Eve's sleep efficiency was highly variable (parent-report range, 47 – 96%; video range, 41 – 84%) during baseline and fell in the clinical range (<75%) at times. Treatment Phase 1 had no effect on sleep efficiency, but Phase 2 had a moderate to large effect (parent-report PEM = 86%, video PEM = 100%). There was further clinically substantive improvement in Phase 3 (parent-report PEM = 95%, video PEM = 100%) which was maintained at follow-up (video PEM = 100%).

Overall sleep quality, CSHQ, and SSR

Parent report, self-report, and video observation showed Eve met criteria for poor sleep quality (as evidenced by reported clinical levels of any sleep variable) on only 25% of nights during Phase 3 and 0% of nights at follow-up, compared with 100% of baseline nights. The total CSHQ score reduced from pre- to post-treatment, however, Eve's score remained within the clinical range (see Table 2). Parent ratings yielded large improvements in the Sleep Onset Delay and Sleep Duration domain scores, though there was little change in other subscale scores. Eve's ratings yielded an improvement in the SSR total score and small improvements in the Sleep Onset Delay, Night Wakings, and Daytime Sleepiness subscale scores (see Table 3). There was no improvement reported in other subscale scores.

Treatment fidelity and child social validity

Treatment fidelity was high but tended to reduce over time. Mean treatment fidelity during Phase 1 was 100%, Phase 2 was 93% (range, 67–100%), and Phase 3 was 70% (range, 60–100%). Mean treatment fidelity during follow-up was 80%. During the post-treatment interview Eve commented on the effectiveness of treatment, noting she could now fall asleep within 30 minutes at bedtime, within 20 minutes during night wakings, and she no longer called out or left her bedroom after bedtime. Eve considered delayed bedtime to be the most helpful and favored treatment component. She enjoyed engaging in progressive muscle relaxation as "it felt nice" but did not find the relaxation resource book helpful. She also disliked not using her e-reader in bed as this had long been part of her bedtime routine. According to the YPTE, Eve considered treatment to be moderately acceptable. YPTE scores have a possible range of 6 to 30, with higher scores indicating higher treatment acceptability. Eve's ratings yielded a total score of 18 (see Supplementary Material Table 8).

Parent social validity

Eve's parents noted the intervention strategies were easy to implement. Eve's mother felt that white noise and stimulus control were critical to intervention success. She reported providing Eve a sense

Table 2. Children's sleep habits questionnaire (CSHQ) pre- and post-treatment scores.

CSHQ scales	Pre	Post
Bedtime Resistance	7	6
Sleep Onset Delay	3	1
Sleep Duration	7	4
Sleep Anxiety	7	6
Night Wakings	3	4
Parasomnias	7	8
Sleep-Disordered Breathing	3	3
Daytime Sleepiness	18	17
Total Score	51	47

Table 3. Sleep self-report (SSR) pre- and post-treatment scores.

SSR Scales	Pre	Post
Bedtime Resistance	4	4
Sleep Onset Delay	2	1
Sleep Duration	2	2
Sleep Anxiety	3	3
Night Wakings	4	3
Daytime Sleepiness	3	2
Total Score	41	36

of control over which sound to choose contributed to her acceptance of the white noise. Eve's mother thought stimulus control increased Eve's sleep pressure and reduced sleep-interfering behavior, as she could not easily access her e-reader during night wakings. The importance of actively including Eve within the therapeutic process was emphasized. Eve's mother noted the communication methods (e.g., letters) were effective in engaging Eve and the incorporation of her interests helped Eve feel "positive" and "excited" about the study. Eve's mother said "not treating her like a subject" by involving her (e.g., explaining treatment rationales) reduced Eve's anxiety regarding the process. She also noted the improvement in Eve's sleep had "affected the whole household"; all family members felt less stressed and irritable, and parent–child interactions were calmer. Additionally, Eve's mother felt the intervention had a positive impact on Eve, who she described as feeling "happier" and "less anxious". She also reported Eve's focus and task completion during the morning routine had improved.

TARF-R scores have a possible range of 17 to 119, with higher scores indicating higher acceptability. Eve's parents' ratings both yielded a score of 100 (see Supplementary Material Table 9). Overall, Eve's parents indicated the intervention package as a whole was effective, reasonable, and low-cost but also rated it as moderately time-consuming and disruptive to their regular routine.

Discussion

In this case study, FBA-informed treatment components were implemented sequentially to address sleep disturbance experienced by a 9-year-old girl with ASD and selective mutism in a minimally sufficient manner. White noise alone had no effect on target sleep variables. White noise and relaxation instruction produced a statistically significant reduction in CCs, an increase in sleep efficiency, as well as a clinically substantive improvement in total sleep duration also. White noise, relaxation instruction, and stimulus control produced statistically significant and clinically substantive improvements across all sleep variables. These improvements were maintained at 10-week follow-up. Further treatment phases involving consequence-based interventions (positive reinforcement and unmodified extinction) were not required. Eve and her parents considered the overall treatment package to be effective, reasonable, and affordable. Their preferred treatment components were white noise and implementation of a faded bedtime.

There is limited evidence for the treatment of sleep disturbance in children with ASD using white noise alone (McLay & France, 2016). Further, while relaxation instruction is commonly incorporated in cognitive behavioral anxiety treatment (Ho et al., 2015) and has been included in multicomponent sleep interventions for young people with ASD (Loring et al., 2016; McCrae et al., 2019; Van Deurs et al., 2019), the efficacy of this technique alone has not been established. Application of individual treatment components alone is unlikely to be able to address the range of antecedent and consequence variables underlying sleep problems. The current study attempted to implement as few and least restrictive components as possible to effectively reduce sleep disturbance. White noise functioned to mask external noises purported to maintain anxiety (e.g., parental discussions) and that were disruptive to sleep (e.g., sibling crying). Relaxation instruction was intended to reduce hyperarousal, lessen the reinforcing value of parent-interaction post-bedtime, and facilitate sleep-conducive behavior. Stimulus control functioned to ensure sufficient homeostatic sleep pressure and strengthen appropriate discriminative stimuli for sleep. While white noise and relaxation instruction had some effect, the combination of white noise, relaxation instruction, and stimulus control were necessary to address the function of Eve's sleep-interfering behavior.

Treatment components were ordered in accordance with behavior function as well as from least to most restrictive and time intensive. Therefore, although lack of physiological sleep pressure was implicated in each of Eve's sleep problems, stimulus control was not introduced until Phase 3. This was because it is more time intensive than white noise or relaxation instruction and can be considered somewhat aversive in that it involves increased parent supervision in the evening (reducing parent alone-time) and meant Eve was not able to use her e-reader in her preferred location. Consequence-

based interventions, including the most restrictive and time-consuming fifth phase (white noise, relaxation instruction, stimulus control, reinforcement, and unmodified extinction), were not required.

This case study shows treatment components can be implemented in a sequential manner to ensure families are not required to engage in numerous unnecessary and/or restrictive strategies. For example, extinction (via removal of parent attention) was not required, although there was a strong attentional component to Eve's sleep disturbance. However, implementation of a multicomponent treatment from the outset may have resulted in faster progress.

In this case, implementation of fewer and less restrictive procedures did not necessarily improve treatment acceptability, compared with TARF-R ratings for comprehensive behavioral sleep interventions involving more restrictive practices (e.g., unmodified or modified extinction; McLay et al., 2017, 2018). Eve's parents still considered treatment to be relatively time-consuming and somewhat disruptive. Eve was resistant to using her bed for sleep only and disliked using her e-reader in an alternative setting. It is common for people on the autism spectrum to become distressed in response to changes in their typical routine. Behavioral sleep interventions by their very nature consist of changes to the child's typical sleep routine and environment. Consequently, less restrictive approaches may still be aversive for family members, particularly in the face of child resistance.

Intervention agents are less likely to comply with treatment procedures as response effort increases (Friman & Poling, 1995). Accordingly, in the current study treatment fidelity reduced during the final treatment phase, which consisted of the most components. Eve and her parents stopped completing relaxation strategies prior to bed and at times Eve was bid goodnight before the agreed faded bedtime. Eve disliked completing relaxation exercises and her parents thought it did not reduce purported hyperarousal. Additionally, the final phase coincided with parental illness. Seemingly, Eve and her parents minimized response effort by only implementing components they preferred and considered to be most effective. Although this phase had a significant treatment effect, video observations showed SOL and duration of NWs still fell within the clinical range on occasion. The reduction in SOL and duration of NWs may have been larger had the treatment plan been followed with more integrity, as was observed during follow-up. This example highlights the importance of composing a treatment plan which includes the fewest, preferred components necessary to produce behavior change, which in turn may enhance treatment fidelity and facilitate maintenance.

Parent-report sleep diary data and video observation indicated there was significant and clinically substantive change in Eve's sleep. This was not reflected in parent- or self-report questionnaire results. Although CSHQ and SSR scores improved over time, the magnitude of difference was relatively small. Further, Eve's total CSHQ score remained in the clinical range of post-treatment. Given the variable number of items within each CSHQ subscale (e.g., Sleep Onset = 1 item, Daytime Sleepiness = 8 items), Johnson et al. (2016) raise concern that the CSHQ may not adequately or reliably measure symptoms of sleep disturbance in ASD. Further, endorsement of specific CSHQ items, such as the child "wets the bed at night", or "is restless and moves a lot during sleep" may be related to the presence of a neurodevelopmental disorder, as opposed to indicative of sleep disturbance (Katz et al., 2018). The CSHQ in its current form (validated on typically developing children) may not be the most effective measure of common sleep concerns for young people with ASD (Johnson et al., 2016). Several studies illustrate revised versions of the CSHQ with four- (Katz et al., 2018) or five-factor models (Johnson et al., 2016; Zaidman-Zait et al., 2020) compared with the original eight-factor structure, may be more appropriate. The newly proposed factors in these studies capture sleep disturbances common among young people with ASD, such as Bedtime Routine problems, Insufficient Sleep and Sleep-onset, as well as Co-sleeping and Sleep Anxiety (Johnson et al., 2016; Katz et al., 2018; Zaidman-Zait et al., 2020). The reliability and validity of these revised factor models require further examination.

Although Pearson product-moment correlations indicated a relatively strong relationship between parent-report and video data, there were large discrepancies in the individual values reported each night (as can be observed in Figures 1–5). This is because Eve's parents were unable to detect the duration of target sleep variables (e.g., SOL, WASO) when she lay quietly in bed and did not seek

them out. As children develop greater autonomy, parents may be less aware of covert sleep disturbances; cognitive and communicative abilities of young people with ASD may inhibit their ability to accurately self-monitor and subsequently report sleep behavior to their parents. In this study, most self-report data did not correlate strongly with video data and IOA between these two measures was low. Eve may have had difficulty remembering numerous brief arousals in the morning. Previous research indicates children and adolescents have difficulty correctly evaluating subjective sleep variables (e.g., duration of NWs; Bauer & Blunden, 2008). Identification of sleep/wake patterns may be even harder for children with ASD (Katz et al., 2018).

An actigraph is a wrist-worn device which uses limb movements as a proxy to measure sleep/wake states and is commonly used to supplement parent-report. Although actigraphy use was considered in the current study, unlike videosomnography, actigraphy cannot reliably identify inactive wake periods or capture salient information (critical to FBA) regarding topographies of sleep and awake behaviors (Katz et al., 2016; Moore et al., 2017). Further it would not likely have provided any additional information than that collected via videosomnography, parent- and self-report sleep diaries, or questionnaires.

A number of limitations should be considered when interpreting the results of this study. Firstly, sequence effects may have impacted treatment outcomes (i.e., white noise, relaxation instruction, and stimulus control may have only been effective due to being preceded by white noise and white noise and relaxation instruction phases). Although results suggest stimulus control was the primary active component within the FBA-informed treatment package, it is not possible to conclude whether stimulus control would have been sufficient alone. Sequence effects were not able to be mitigated by employing reversal conditions between phases, given relaxation instruction resulted in skill improvement which could not be eliminated. Secondly, as follow-up data were collected only once at 10 weeks post-treatment, the long-term maintenance of this intervention was not established. Thirdly, while a minimally sufficient approach was effective in eliminating sleep disturbance for the participant within this study, this approach may not be appropriate or effective for other children on the autism spectrum with diverse sleep presentations and varying communicative abilities. To increase generalizability to young people across the autism spectrum, the relationship between participant communicative abilities, cognitive functioning, and treatment outcomes warrants further investigation. Replication with larger, diverse samples whereby sequence effects are accounted for is necessary to demonstrate the efficacy of minimally sufficient FBA-informed sleep interventions. Future research could implement treatment components individually to evaluate the necessity and sufficiency of each component alone, before combining components into a treatment package. This is critical to identify active treatment components and ensure clinicians and families can implement the least restrictive and minimally sufficient interventions necessary to treat sleep problems, improving social validity and treatment maintenance.

Author contributions

JvD: designed and executed the study, conducted data analyses, and wrote the paper. KF and LM contributed equally towards the collaboration of the design, execution, and writing of the study. NB: collaborated with the data analysis, writing of the study, and constructed the figures.

Compliance with ethical standards

All procedures performed in studies involving human participants were in accordance with the ethical standards of the University of Canterbury Human Ethics Committee and with the Code of Ethics for Psychologists Working in Aotearoa New Zealand.

Disclosure statement

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Informed consent

Eve's caregivers provided written informed consent and Eve provided written assent prior to inclusion in the study.

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